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The Journal of
Bone and Joint Surgery

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EDITORIALS AND ANNOTATIONS

TUBERCULOSIS OF THE SPINE

Most orthopaedic surgeons in this country were brought up to believe that it was wicked to perform a planned operative exposure of an uncontaminated tuberculous focus: a little over ten years ago the surgeon who opened a "cold abscess" of the spine would usually experience an acute sense of guilt. Before streptomycin was available such operations involved very considerable risks; nevertheless, there were brilliant exceptions and it is important to remember that the classical researches of Capener (1954) and Dott (1947) and Alexander (1946) on Pott's paraplegia were done before the advent of antibiotics.

In the last decade many surgeons have been operating on paravertebral abscesses and tuberculous foci in the spine (Wilkinson 1954, 1955; Roaf 1954; Fellander 1955). During the last few years in many centres the tendency has been to be increasingly daring. In this time we have learned four things. Firstly, immediately after operation there is dramatic relief of pain and a striking improvement in the patient's general condition and sense of well-being. Secondly, it is rare not to find bony sequestra, necrotic disc material, caseous matter, inspissated pus and other debris which one would certainly consider a bar to healing in non-tuberculous chronic osteomyelitis. Thirdly, even after prolonged conservative treatment—including antibiotics and chemotherapy—living mycobacteria can often be found in the centre of the avascular focus, apparently protected by scar tissue from blood-borne therapeutic agents. Lastly, we have learned how deceptive the radiographic appearances usually are. A paravertebral shadow may contain highly infective pus or merely consist of fibrous tissue; an apparently well healed spinal lesion, confidently alleged to be the site of bony ankylosis, is often a smouldering focus ready to burst out at the slightest provocation.

However, in case the "interventionists" become unduly complacent we must remind ourselves of the many things we do not know. It has not yet been shown that operation prevents late recurrence and increases the chance of permanent cure; so far, the follow-up has been short and there has been no true comparison of like with like. The natural history of tuberculosis of the spine varies in different parts of the world, with the anatomical site and with the patient's age and condition. A surgeon who claims that his percentage of "cures" in 1956 is higher than those of another surgeon working in a different part of the world in 1950 does not necessarily prove that his methods are sounder. Even if operation is advantageous we do not know whether such intervention should be early or late. Nor do we know what length of conservative treatment is desirable or what is the best combination of antibiotic and chemotherapeutic agents, bearing in mind the psychological, educational, domestic, social and economic factors that have to be considered before advising a lengthy stay in hospital or a long course of expensive drugs.

Even in the realm of surgical technique there is much that is unknown. For access to the cervical region, the surgical world is divided into three camps—in front of, behind, or through, the sternomastoid (Southwick and Robinson 1957). In the cervico-thoracic region a direct, trans-sternal approach, with division of the innominate vein, has been advocated

(Cauchoix and Binet 1957); others prefer the more oblique but less formidable cervical rib approach. In the thoracic region the "trans-pleuralists" (Hodgson and Stock 1956) claim better access, while the "extra-pleuralists" (Jones 1958) believe that they have fewer anaesthetic problems and a smoother convalescence. In the lumbar region an extra-peritoneal approach is favoured by most surgeons. The extent of intervention also varies. Some merely drain the abscess on one side. Others follow the Russian example and excise the whole of the abscess and abscess wall (Kirillov and Kon 1956). Many believe that a thorough cleansing of the bony focus is essential; some extend their intervention to the contralateral abscess—at the same or another session; the bolder spirits pack cancellous bone into the residual cavity. Some surgeons insert a tube for local instillation of streptomycin: others abhor this. These operations can be difficult and, whatever approach is used, accurate radiological localisation of the lesion before and, often, during the operation is essential. It is important to repeat that we do not yet know which is the best procedure under any given circumstance. The fact remains that spinal tuberculosis, especially in the thoracic and cervical spine, is still a dangerous and relentless condition (Dobson 1951; Scott 1955).

Some may feel that the subject hardly merits much time and thought. The incidence of spinal tuberculosis in this country is falling; long-stay beds are easily available; there is no shortage of anti-tuberculous drugs; the Welfare State has removed many of the problems of a prolonged sojourn in hospital. Such an attitude is completely wrong and disastrously parochial. In less developed countries tuberculosis of bones and joints is still a major economic problem: hospital beds are few, drugs are scarce and expensive, the economic and social consequences of prolonged invalidism are disastrous (Konstam and Konstam 1958). It is an urgent necessity to know how best to use what slender resources there are. Five years ago, under difficult circumstances, the working rule was adopted of "no surgery without streptomycin, no streptomycin without surgery." This slogan appeared to be true on a short-term basis, but we still do not really know the long-term results. It is a reproach to orthopaedic surgery that we have not been able to institute an investigation into the treatment of skeletal tuberculosis comparable to the investigation into the treatment of pulmonary tuberculosis now being undertaken in Madras under the auspices of the Medical Research Councils of the United Kingdom and India.

Finally, to repeat a platitude, tuberculosis of the spine is a local manifestation of a systemic infection. However much surgical treatment of the spinal focus may appear to lead to quicker local healing, it does not by itself cure the systemic infection. If we neglect this it is probable that another focus will develop later. ROBERT ROAF.

REFERENCES

ALEXANDER, G. L. (1946): Neurological complications of Spinal Tuberculosis. Proceedings of the Royal Society of Medicine (Section of Neurology with Section of Orthopaedics), **39,** 730.

CAPENER, N. (1954): The Evolution of Lateral Rhachotomy. Journal of Bone and Joint Surgery, **36–B,** 173.

CAUCHOIX, J., and BINET, J. P. (1957): Anterior Surgical Approaches to the Spine. Annals of the Royal College of Surgeons of England, **21,** 237.

DOBSON, J. (1951): Tuberculosis of the Spine. Journal of Bone and Joint Surgery, **33–B,** 517.

DOTT, N. M. (1947): Skeletal Traction and Anterior Decompression in the Management of Pott's Paraplegia. Edinburgh Medical Journal, **54,** 620.

FELLANDER, M. R. (1955): Radical Operation in Tuberculosis of the Spine. Acta Orthopaedica Scandinavica, Supplement No. 19.

HODGSON, A. R., and STOCK, F. E. (1956): Anterior Spinal Fusion. British Journal of Surgery, **44,** 266.

JONES, B. S. (1958): Pott's Paraplegia in the Nigerian. Journal of Bone and Joint Surgery, **40–B,** 16.

KIRILLOV, B. P., and KON, I. I. (1956): Khirurgicheskoe lechenie natechnykh abstsessov pri tuberkuleznom spondilite. [Surgical treatment of gravity abscesses in tuberculous spondylitis.] Khirurgiia, Moscow, **32,** 53.

KONSTAM, P. G., and KONSTAM, S. T. (1958): Spinal Tuberculosis in Southern Nigeria. Journal of Bone and Joint Surgery, **40–B,** 26.

ROAF, R. (1954): The Surgical Treatment of Skeletal Tuberculosis. Indian Journal of Surgery, **16,** 216.
SCOTT, J. C. (1955): Treatment of Tuberculosis of the Spine. Journal of Bone and Joint Surgery, **37–B,** 367.
SOUTHWICK, W. O., and ROBINSON, R. A. (1957): Surgical Approaches to the Vertebral Bodies in the Cervical and Lumbar Regions. Journal of Bone and Joint Surgery, **39–A,** 631.
WILKINSON, M. C. (1954): Chemotherapy of Tuberculosis of Bones and Joints. Journal of Bone and Joint Surgery, **36–B,** 23.
WILKINSON, M. C. (1955): The Treatment of Tuberculosis of the Spine by Evacuation of the Paravertebral Abscess and Curettage of the Vertebral Bodies. Journal of Bone and Joint Surgery, **37–B,** 382.

TUBERCULOSIS OF THE PUBIS

Report of Eleven Cases

O. R. NICHOLSON, AUCKLAND, NEW ZEALAND

From the Institute of Orthopaedics, Royal National Orthopaedic Hospital, London

Tuberculosis of the pubis is uncommon. It is not mentioned in text-books on skeletal tuberculosis by Girdlestone and Somerville (1952) or Sanchis-Olmos (1948), but a full account was given by Sorrel and Sorrel-Déjerine (1932).

Since the first reports by Thilesen (1855) and by Hennies (1888) over 150 cases have been recorded, the first in English being by Jackson (1923). A further fifteen appear in the English literature (Clairmont 1925, Bean 1930, Alpert 1939, Bevan 1955, Fairbank 1955, Read 1955) and Continental sources contain several more (Pytel 1935, Greger 1938, Ficai 1949, Clavel 1951). The largest series is that of Sorrel and Sorrel-Déjerine, with twenty-six cases.

The eleven patients described here were seen between 1937 and 1954, five at the Royal National Orthopaedic Hospital, London and Stanmore, and six at Middlemore Hospital, Auckland. They form the largest group so far reported in English.

CASE REPORTS

Case 1—A boy aged seven years was admitted in 1953. For about six weeks he had complained of pain in the right groin and limped. There was diffuse thickening over the symphysis but no palpable abscess. Radiographs showed extensive erosion of the body of the right pubis (Fig. 1).

FIG. 1
Case 1—Boy aged seven years. Erosion of body of right pubis.

Investigation and treatment—An inguinal gland biopsy was positive for tuberculosis. He was kept in bed, and streptomycin (0·75 gramme daily by intramuscular injection), para-aminosalicylic acid (8 grammes daily by mouth) and iso-nicotinic-acid-hydrazide (100 milligrams daily by mouth) were

given for three months. A high-tone deafness developed, and para-aminosalicylic acid and iso-nicotinic-acid-hydrazide were given alone for a further three months.
Progress—After seven months he was discharged on crutches but within one month pain had recurred

FIG. 2
Case 1—Two and a half years later. There is slight upward subluxation of the right half of the pelvis.

and he was readmitted. Radiographs showed calcified debris on the right and rarefaction of the body of the left pubis. An abscess was present above the symphysis pubis, and although this was aspirated on four occasions it continued to re-form.
Operation—A month later the affected area was curetted and the wound was closed. A small sinus persisted for two months. After the operation para-aminosalicylic acid (8 grammes daily by mouth) and iso-nicotinic-acid-hydrazide (100 milligrams daily by mouth) were given for three months and the patient was discharged.
Further progress—Three years later he was symptomless and leading a normal life. Radiographs showed no evidence of activity but slight upward displacement of the right half of the pelvis (Fig. 2). No other tuberculous lesion was found.

Case 2—A man aged nineteen years was admitted in 1941 with a swelling in the left thigh. Five years before he had been treated for pulmonary tuberculosis, and two years later had been in hospital for a year for treatment of a swelling over the symphysis pubis. Aspiration had shown it to contain tuberculous pus.

Radiographs showed erosion of the body of the left pubis extending laterally from the symphysis.
Treatment—The swelling in the left thigh was aspirated, yielding tuberculous pus. The abscess did not re-form, and after eight months' conservative treatment the symphysis was fused by a rib graft. No abscess was found.
Progress—The wound healed, and he was discharged after two months and resumed work after three months. Seven years later the patient was symptomless.

Case 3—A man aged eighteen years was admitted in 1953. He had noticed pain in the groin for six months after a fall when both legs were forcibly abducted. Three weeks before admission he had noticed a swelling on the medial aspect of the right thigh. Radiographs showed almost symmetrical erosion of the bodies of both pubic bones without sequestrum formation (Fig. 3).
Treatment—The abscess was incised and found to track deep to the adductors, and also superficially up to the symphysis pubis. Through a suprapubic incision the affected area was curetted. The suprapubic incision was closed and the abscess cavity packed. Tubercle bacilli were isolated from the pus.
Progress—The suprapubic wound healed but a sinus in the adductor region persisted for five months. After the operation streptomycin (1 gramme daily by intramuscular injection) and para-aminosalicylic

acid (15 grammes daily by mouth) were given for three months. An ischio-rectal abscess developed but soon settled after incision. Tubercle bacilli could not be isolated from it.

A year later the patient was symptomless even when doing heavy work. Radiographs showed no evidence of activity (Fig. 4). No other tuberculous lesion was found.

FIG. 3

Case 3—Youth aged eighteen years. Almost symmetrical erosion of both pubic bones

FIG. 4

Case 3—Nine months after drainage of the abscess and curettage of the lesion. Lesion quiescent.

Case 4—A man aged twenty-two years was admitted in 1948. Seven months before he had developed an aching pain in the suprapubic region, and later noticed a lump. This had increased in size until it had discharged pus through a sinus above the symphysis pubis. The patient had previously been treated for pulmonary tuberculosis and for tuberculosis of the left elbow. Radiographs showed a

FIG. 5
Case 4—Man aged twenty-two years. Extensive destruction of body of pubis with large sequestrum.

FIG. 6
Case 4—Sinograph showing widespread ramifications of the abscess cavity.

FIG. 7
Case 4—Six years later. The lesion has healed but during the period of activity both pubic bones have become involved.

destructive lesion of the body of the left pubis and a large sequestrum (Fig. 5). Injection of the sinus with lipiodol showed the ramifications of the abscess (Fig. 6).

Treatment—One month later the abscess was opened, the dense scar tissue of the wall was excised and the sequestra were removed. The cavity was curetted and packed.

Progress—The wound healed slowly, a sinus persisting for twelve months. Five years later the patient was symptomless and was doing heavy labouring work. Radiographs showed that the infection had at some stage also involved the body of the right pubis, but there was no evidence of activity in either lesion (Fig. 7).

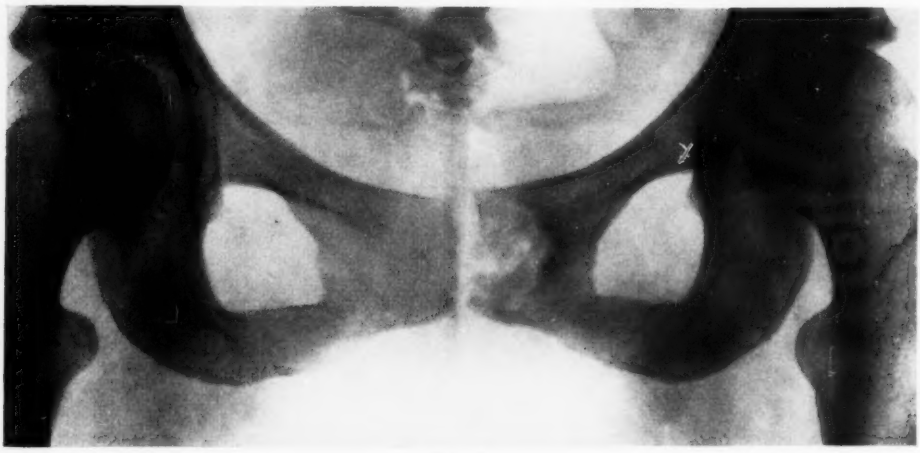

FIG. 8

Case 5—Woman aged twenty-six years. Irregular erosion of body of left pubis and rarefaction of right pubis.

FIG. 9

Case 5—Six years later. The symphysis is bridged by bone.

Case 5—A woman aged twenty-six years was seen in 1949. She had noticed pain in the pubic region for four months and this had been worse on standing. Radiographs showed irregular erosion of the left pubis and rarefaction extending laterally (Fig. 8). Two months later an abscess developed in the left labium.

Treatment—The abscess was aspirated and tubercle bacilli were isolated. After this a sinus persisted and about a month later the affected area was curetted and the wound closed. Streptomycin (1 gramme daily by intramuscular injection) was given for five weeks.

Progress—The wound healed and the sinus closed. The patient was discharged after six weeks but

three months later again developed pain, and six months after the operation she was readmitted. Radiographs showed that the body of the right pubis was also affected. The affected area was again curetted. The wound healed and the patient was discharged after two months. Streptomycin (1 gramme daily by intramuscular injection) and para-aminosalicylic acid (15 grammes daily by mouth) were given for three months after the operation.

Three years later the patient was symptomless, and radiographs showed bone bridging the symphysis. A year later she had a normal delivery of a full-term infant. Radiographs in 1955 showed bone trabeculation across the symphysis (Fig. 9). No other tuberculous lesion was found.

Case 6—A man aged twenty-four years was admitted in 1950. Two weeks before he had developed an aching pain in the left groin while working and had noticed a swelling. The pain became acute and he was unable to walk. On examination there was an abscess in the left groin and tenderness over the symphysis pubis. Radiographs showed a destructive lesion involving the bodies of both pubic bones without sequestrum formation.

Treatment—After rest in bed and leg traction for a month the abscess was opened and the affected area curetted. Tubercle bacilli were isolated from the pus. The wound healed and the patient was discharged after a month.

Progress—Six months later he was symptomless and there was no recurrence of the abscess. Radiographs suggested that the lesion was becoming quiescent. No other tuberculous lesion was found.

Case 7—A woman aged twenty-nine years was admitted in 1951. For several months she had noticed a nagging pain in the right groin, a feeling of weakness in the leg and a limp. Shortly before admission a swelling appeared in the right groin and extended into the labium majus. Aspiration had shown tuberculous pus. Radiographs showed a destructive lesion of the bodies of both pubic bones with sequestrum formation.

Treatment and progress—After rest in bed for two months and a course of streptomycin, the affected area was curetted and the sequestrum removed. The wound healed. Three years later the patient had no symptoms from the pubis and there was no recurrence of the abscess, but she was suffering from lupus erythematosus. Radiographs showed no evidence of activity.

Case 8—A man aged thirty years was admitted in 1937. Two years before he had been treated for tuberculous peritonitis. For eight months a swelling had been noticed in the left groin and for five months another swelling on the inner aspect of the left thigh. Three months before admission aspiration of the swelling in the groin had shown tuberculous pus. A sinus had formed and was still present. Radiographs showed irregular erosion of the bodies of both pubic bones, especially the left, with sclerosis at the margins. No sequestrum was present.

Treatment and progress—Operation was undertaken three days after admission. Through a suprapubic incision the affected area was curetted and swabbed with ether. The wound was closed. It healed but the sinus persisted. The discharge was much reduced after a second operation, two months after the first, when a small sequestrum was removed. Two years later he was symptomless but a small sinus persisted. Radiographs showed no evidence of activity.

Case 9—A woman aged thirty-six years was transferred from a sanatorium in 1949. She was under treatment for pulmonary tuberculosis and had developed a painless swelling above the symphysis pubis about one year before which had gradually increased in size. Radiographs showed irregular erosion of the bodies of both pubic bones, especially the right, and several small sequestra.

Treatment and progress—At operation a month after admission the swelling was incised and found to contain tuberculous pus. The abscess extended upwards from the symphysis pubis beneath the rectus abdominis muscle. The wound was packed. The patient was nursed on a plaster bed for six months, streptomycin (1 gramme daily by intramuscular injection) being given for the first month after operation. Three years later the patient was symptomless and there was no recurrence of the abscess. Radiographs showed no evidence of activity.

Case 10—A woman aged sixty-eight years was admitted in 1954. For about five months she had complained of a severe aching pain in the left groin when walking. This had increased in severity and she had been unable to walk for two weeks. She had not noticed any swelling. On examination there was a diffuse tender swelling above the symphysis. The patient had a marked limp and was unable to stand on the left leg because of pain. Radiographs showed a destructive lesion of the bodies of both pubic bones, with several small sequestra.

Treatment and progress—After two weeks' rest in bed, with streptomycin (1 gramme daily by intramuscular injection) and para-aminosalicylic acid (15 grammes daily by mouth), the whole of the affected area was excised, about half an inch of bone on each side of the symphysis being removed. The wound discharged for three weeks and then healed. Histological examination of the material removed at operation showed the appearances of tuberculosis. The patient was discharged eight weeks after operation and a year later she was symptomless.

FIG. 10

Case 11—Man aged fifty-five years. Involvement of one pubis with formation of a small sequestrum. In this case the lesion remained unilateral.

Case 11—A man aged fifty-five years was admitted in 1949. He had noticed tenderness over the symphysis pubis for six months and had first noticed a swelling after slipping and striking the groin three months previously. Radiographs taken before his admission three months after the onset of symptoms showed erosion of the body of the left pubis with a small sequestrum (Fig. 10). Radiographs on admission showed further erosion.

Treatment and progress—The swelling was aspirated and was found to contain tuberculous pus. After four months' rest in bed the abscess was still present, so it was incised and packed. Healing was slow, a sinus persisting for twelve months. Three years later the patient was symptomless and had no recurrence of the abscess. Radiographs showed no evidence of activity and the lesion had remained unilateral. No other tuberculous lesion was found.

DISCUSSION

Clinical features. *Age of onset*—The ages of the patients in this group range from seven to sixty-eight years. Greger (1938) found reports of the disease in patients of from three to seventy years of age. Sorrel and Sorrel-Déjerine (1932) found that although the condition might occur at any age, it was rare in adults. It appears, however, that no age is immune or particularly susceptible.

Symptoms and signs—The insidious onset of symptoms is commented on by all previous authors, but the history is not necessarily long. In the present series it varied from two weeks to one year. Seven of the eleven patients complained of an aching pain in the groin, and in two cases this became severe enough to keep them in bed. Some irregularity of gait was commonly present because of the increased discomfort on weight bearing, but no patient complained of pain in the region of the sacro-iliac joint as might be expected if there was instability of the pelvis. The patient described by Bean (1930) complained of sacro-iliac pain, and backache as a symptom was mentioned by Alpert (1939).

The pain is less severe than that of non-suppurative osteitis pubis, and unlike the pain of that condition it is not usually severe when the patient is at rest. Local tenderness over the symphysis was a constant feature.

Abscess formation was present in nine of the eleven cases. The abscess may present above the symphysis, in the groin or in the thigh. Sinus formation had occurred only once before the patient was seen, and in this case the ramifications of the abscess cavity were

demonstrated after the injection of lipiodol (Fig. 6). The abscess was often found to be extensive at operation. There seems little support for the statement of Bevan (1955) that abscess formation is uncommon.

Tuberculous lesions elsewhere—Lesions elsewhere were apparent in only four of the eleven cases, and it has been noted by previous authors that this might be the only demonstrable focus.

Relationship to injury—Injury to the symphysis has been incriminated for all the conditions that affect that region, and a case of staphylococcal osteomyelitis was reported by Adams and Chandler (1953) as one of post-traumatic osteitis pubis. In Case 3 of this series the onset of symptoms certainly followed a local injury, and in Case 11 symptoms had begun three months after an injury. It may be that local injury determines the site of the lesion in patients who at the time have a tuberculous bacillaemia.

Incidence—Tuberculosis of the symphysis pubis is certainly uncommon, but it is likely that a number of cases are not correctly diagnosed. Sorrel and Sorrel-Déjerine (1932) had seen more cases of tuberculosis of the symphysis (twenty cases between 1920–30 and six cases before that) than of tuberculosis of the greater trochanter, and Read (1955) reported seeing four cases in eight years. Six of the present series occurred in seven years in an area in New Zealand where skeletal tuberculosis is not particularly common.

Radiological features—Erosion of the body of the pubis is constant, as also is some degree of rarefaction of the adjacent rami. The rarefaction is not as extreme as in osteitis pubis. The erosion may be unilateral or bilateral, and sometimes symmetrical, but it appears to start in the body of the pubis adjacent to the symphysis pubis and then to spread across the symphysis. Wheeler (1941), referring to osteitis pubis, stated that " it is inconceivable that infection would jump over the cartilaginous symphysis extending to an equal distance bilaterally," and Mortensen (1951), who is one of the few authors to mention tuberculosis in the differential diagnosis of lesions of the pubis, stated that the destruction is predominantly unilateral. This is, however, certainly not invariable.

Sequestration occurred in six cases, and in the remainder tuberculous debris was present. Sequestration is rare in osteitis pubis (Stutter 1954) but does occur when there is actual infection of the pubic bones.

Differential diagnosis. *Infections of the pubis*—Staphylococcal osteomyelitis (Dinan 1939) and osteomyelitis from bacillus coli, pseudomonas aeroginisa and bacillus proteus have been described (Wilensky 1938, Abrams, Sedlezky and Stearns 1949; Lavalle and Hamm 1951) and may occur acutely or insidiously. The case of post-traumatic osteitis pubis described by Adams and Chandler (1953) seems to be of this group and not a separate entity. The diagnosis of these conditions depends on the isolation of the organisms from the pus if an abscess is present. The presence of a tuberculous lesion elsewhere favours the diagnosis of tuberculosis of the pubis, but the radiological appearances are not diagnostic.

Osteitis pubis—This term is now generally restricted to those cases which occur in men after suprapubic operations (usually prostatectomy) and in women after pregnancy, pelvic operations and trauma to the symphysis (Golden 1952, Wiltse and Frantz 1956). These cases are characterised by intense pain, but abscess formation does not occur. Radiographs show marked rarefaction of the pelvis on both sides of the symphysis pubis; sequestration is rare. The condition is usually ascribed to some vascular disturbance and may be associated with retropubic infection (Stutter 1954). Cases in which there is actual infection of the symphysis pubis have in the past been included in this group but comprise a separate entity.

Adolescent osteochondritis of the symphysis pubis (Burman, Weinkle and Langsam 1934)—This is an uncommon condition in which local pain and tenderness occur without abscess formation. Radiographs show irregularity of the margins of the symphysis pubis.

Treatment—Thilesen (1855) and Hennies (1888) successfully treated their patients by curettage. Sorrel and Sorrel-Déjerine (1932) advocated a direct attack on the lesion and stated that

sinuses will then close spontaneously. Radical excision is not required, though Clairmont (1925) stated that, in his experience, a wide excision of the soft parts and of the pubis was necessary.

Operation was carried out in all cases in the present series. In one patient the whole of the affected bone was excised (Case 10), and in another (Case 2) a bone graft was placed across the symphysis pubis. In the remaining nine patients the lesion was curetted and if an abscess was present it was drained.

Aspiration of the abscess would be indicated as a diagnostic measure or if there were danger of the skin's breaking down.

Bone grafting was carried out in only one case. Bean (1930), believing that his patient had osteitis fibrosa cystica, opened into an abscess cavity and then placed a tibial bone graft across the symphysis pubis. The graft subsequently sequestrated but the patient, who before operation had complained of sacro-iliac pain, lost his symptoms. Alpert (1939) believed that bilateral sacro-iliac fusion should be carried out in order to maintain stability of the pelvic ring. This was successful in one of his patients and failed in another who, however, was cured by curettage of the lesion and remained symptomless.

In none of the cases reported here did symptoms of pelvic instability develop after operation without bone grafting.

It has been stated (Greger 1938) that there would be little chance of parturition if the symphysis were fused. Parturition did, however, take place normally in Case 5, in which solid bony fusion occurred after two curettings.

There was little systemic disturbance in any of the patients, and, although periods of rest in bed were used in several cases, this does not seem necessary.

Antibiotics were used in six of the eleven cases; healing was complete in all these cases by the end of six months, and in four by the end of two months. In the patients treated without antibiotics, sinuses persisted for three years in one case (Case 8) and for one year in two cases (Cases 4 and 11). It is suggested that, in the absence of lesions elsewhere requiring more prolonged treatment, antibiotics be used only until the wound has healed.

Results—The duration of follow-up of the patients reported here varied from one to seven years, the average being three years. In all cases the lesion responded satisfactorily to treatment and the course of the disease did not seem to be particularly influenced by the use of antibiotics or by the presence of tuberculous lesions elsewhere.

The disease as seen in the patients in New Zealand does not seem to differ from that seen in England.

SUMMARY

1. Eleven cases of tuberculosis of the pubis are described.

2. The disease is often of insidious onset, and symptoms vary from vague discomfort to incapacitating pain in the region of the symphysis and the groin.

3. Abscess formation is common and was present in nine of the eleven patients when they first attended.

4. The lesion has a good prognosis and responds well to simple curettage.

5. In this series operation, without bone grafting, has not been followed by pelvic instability or back pain.

I wish to thank the staffs of the Royal National Orthopaedic Hospital, London and Stanmore, and of Middlemore Hospital, Auckland, New Zealand, for permission to review the case notes of patients admitted under their care. Cases 3, 4, 6, 7, 10 and 11 were presented at the 1955 meeting of the New Zealand Orthopaedic Association by Mr J. R. Kirker.

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REFERENCES

ABRAMS, M., SEDLEZKY, I., and STEARNS, D. B. (1949): Osteitis Pubis. New England Journal of Medicine, **240,** 637.

ADAMS, R. J., and CHANDLER, F. A. (1953): Osteitis Pubis of Traumatic Etiology. Journal of Bone and Joint Surgery, **35–A,** 685.

ALPERT, L. (1939): Tuberculosis of the Symphysis Pubis. New England Journal of Medicine, **220,** 786.

BEAN, H. C. (1930): Tuberculosis of the Symphysis Pubis. Journal of Bone and Joint Surgery, **12,** 345.

BEVAN, P. G. (1955): Tuberculosis of Pubis Presenting as a Cold Abscess of the Thigh. British Medical Journal, **ii,** 832.

BURMAN, M., WEINKLE, I. N., and LANGSAM, M. J. (1934): Adolescent Osteochondritis of the Symphysis Pubis. With a Consideration of the Normal Roentgenographic Changes in the Symphysis Pubis. Journal of Bone and Joint Surgery, **16,** 649.

CLAIRMONT, P. J. (1925): Tuberculous Osteitis of Symphysis Pubis, with Sinus Formation and Mixed Infection with the Staphylococcus Aureus. British Journal of Surgery, **12,** 606.

CLAVEL, M. (1951): Tres Casos de Tuberculosis del Pubis. Cirugía del Aparato Locomotor, **8,** 149.

DINAN, J. J. (1939): Two Cases of Primary Osteomyelitis of the Pubic Bone. Canadian Medical Association Journal, **41,** 436.

FAIRBANK, H. A. T. (1955): Tuberculosis of the Pubis. (Correspondence) British Medical Journal, **ii,** 1,028.

FICAI, A. (1949): Due Casi di Tubercolosi del Pube con deflusso ascessuale non comune. Minerva Chirurgica, **4,** 457.

GIRDLESTONE, G. R., and SOMERVILLE, E. W. (1952): Tuberculosis of Bone and Joint. Second edition. London, New York, Toronto: Geoffrey Cumberlege, Oxford University Press.

GOLDEN, A. (1952): Lesions of Ischium and Pubis in Pregnancy Resembling Osteitis. Journal of Urology, **67,** 370.

GREGER, E. (1938): Five Cases of Tuberculosis of the Symphysis Pubis. Acta Orthopaedica Scandinavica, **9,** 43.

HENNIES, E. (1888): Zur Kenntnis der Fälle von tuberkulöser Caries der Symphyse auf Grund klinischer Beobachtung. (Inaugural-Dissertation) Griefswald. (Quoted by Jackson and by Bean.)

JACKSON, J. B. (1923): A Case of Tuberculosis of the Symphysis Pubis. American Journal of Roentgenology, **10,** 806.

KIRKER, J. R. (1956): Tuberculous Osteitis Pubis. Journal of Bone and Joint Surgery, **38–B,** 602.

LAVALLE, L. L., and HAMM, F. C. (1951): Osteitis Pubis: its Etiology and Pathology. Journal of Urology, **66,** 418.

MORTENSEN, H. (1951): Osteitis Pubis. Australian and New Zealand Journal of Surgery, **21,** 103.

PYTEL, A. (1935): Tuberculose de l'Articulation pubienne. Revue d'Orthopédie, **22,** 348.

READ, R. F. (1955): Tuberculosis of the Pubis. (Correspondence.) Lancet, **ii,** 1,140.

SANCHÍS-OLMOS, V. (1948): Skeletal Tuberculosis. Baltimore: The Williams and Wilkins Company.

SORREL, E., and SORREL-DÉJERINE, Mme (1932): Tuberculose osseuse et ostéo-articulaire. Paris: Masson et Cie.

STUTTER, B. D. (1954): The Complications of " Osteitis pubis." British Journal of Surgery, **42,** 164.

THILESEN (1855): Tilfaelde av necrose i bekkenbenene, stendannelse om et i blaeren indtraengt necrotisert benstykke etc. Norsk Magasin for Laegevidenskapen (quoted by Greger).

WHEELER, W. K. (1941): Periostitis Pubis Following Suprapubic Cystostomy. Journal of Urology, **45,** 467.

WILENSKY, A. O. (1938): Osteomyelitis of the Pelvic Girdle. Archives of Surgery, **37,** 371.

WILTSE, L. L., and FRANTZ, C. H. (1956): Non-suppurative Osteitis Pubis in the Female. Journal of Bone and Joint Surgery, **38–A,** 500.

POTT'S PARAPLEGIA IN THE NIGERIAN

B. S. JONES, CAPE TOWN, SOUTH AFRICA

From the Igbobi Orthopaedic Hospital, Lagos, Nigeria

Although most victims of Pott's paraplegia recover completely with conservative treatment alone, the tragedy of that small but significant percentage that remain permanently paralysed still presents a challenge.

The operation of antero-lateral decompression has earned a permanent place in the management of these cases, and Seddon (1956) has recently defined the indications for its employment, a fitting sequel to the classical papers in which he (1935) and Butler (1935) did so much to clarify the pathogenesis and prognosis of the condition.

The purpose of this paper is to review the cases of Pott's paraplegia treated in this hospital during six and a half years, with particular reference to the place of operative intervention.

CLINICAL MATERIAL AND GENERAL DATA

There were admitted to the Igbobi Orthopaedic Hospital, Lagos, between January 1949 and June 1955, 276 patients with tuberculosis of the spine, of whom 115 at one time or another suffered paraplegia or paraparesis. This unusually high percentage was due to a shortage of beds, which necessitated the treatment of most patients with uncomplicated tuberculous spinal disease as out-patients. All the patients were African. Of the paraplegic patients sixty were male and fifty-five female.

Eight patients died while in hospital, three from pressure sores and toxaemia, three from tuberculous meningitis, one from cerebral malaria and one from an unknown cause. Eighty-six patients recovered completely; ten recovered with minor residual effects; eleven were permanently paralysed, seven partially and four totally.

Age—The highest quinquennial incidence was at five to nine, and the second highest ten to fourteen (Table I).

Vertebral bodies affected—The sixth, seventh, eighth and ninth were the thoracic vertebrae affected most often (Table II). The average number involved in patients with paraplegia was 3·5, in cases without paraplegia 3·4 (Table III).

Grade of paraplegia according to onset—The distribution of cases according to grade of paraplegia is shown in Table IV, grade 1 representing a spastic paraplegia in which some voluntary control is retained, grade 2 a severe paraplegia in extension with complete loss of voluntary movement, grade 3 a paraplegia in flexion and grade 4 a flaccid paraplegia.

GRADES OF PARAPLEGIA

EARLY ONSET PARAPLEGIA

Grade 1—There were twenty-five cases of early onset paraplegia of grade 1, of which nineteen persisted unchanged for up to six months, three for six to twelve months, one for twelve to twenty-four months, two for over twenty-four months. Twenty were treated by purely conservative means—that is, on alternated posterior and anterior plaster shells with (in all but the earliest cases) streptomycin twice weekly, supplemented by para-amino-salicylic acid alone at first and by sulphaniazid also in later cases for a course of three to six months.

Costo-transversectomy was undertaken in four patients and in one a paravertebral abscess was aspirated. Twenty-three patients recovered completely. One patient with a three years' history and with a severe kyphos and an apparently healed spinal lesion remained permanently

TABLE I

AGE (YEARS)

0–4	5–9	10–14	15–19	20–29	30–39	40 and over
22	40	32	10	5	4	2

TABLE II

DISTRIBUTION

Vertebrae involved	C	T.1	T.2	T.3	T.4	T.5	T.6	T.7	T.8	T.9	T.10	T.11	T.12	L.1	L.2	L.3
Frequency . .	3	6	11	22	28	38	46	49	49	43	37	30	20	13	6	4

TABLE III

AVERAGE NUMBER OF VERTEBRAE INVOLVED

Paraplegic cases	Non-paraplegic cases
3·5	3·4

TABLE IV

TYPE AND GRADE OF PARAPLEGIA

	Grade 1	Grade 2	Grade 3	Grade 4	Total
Early onset	25	56	11	10	102
Late onset	4	5	3	1	13
			Grand total		115

TABLE V

CORRELATION BETWEEN PARAPLEGIA AND STATE OF ABSCESS

	Bone lesion active	Lesion apparently inactive	Paravertebral abscess	Recovery of paraplegia coincident with absorption of abscess
Early onset	100	2	88	44
Late onset	8	5	7	5

TABLE VI

DURATION OF FOLLOW-UP

Follow-up	5 years and over	4 years	3 years	2 years	1 year	Less than 1 year
Cases	9	10	26	26	28	16

paraplegic. Another patient with disease of almost three years' duration with an apparently healed spinal lesion but a persistent paravertebral abscess shadow and anterior scalloping of the vertebral bodies was treated by costo-transversectomy. Eighty cubic centimetres of inspissated pus were found retained beneath the anterior longitudinal ligament, and almost full recovery followed its release.

Grade 2—There were fifty-six patients with early onset paraplegia of grade 2, of whom forty-six recovered completely and ten did not. Of these ten, four (three treated entirely conservatively and one by an unproductive costo-transversectomy) recovered with only trifling residual paresis. Three, one subjected to an unproductive costo-transversectomy after three months, one to an unproductive costo-transversectomy and laminectomy after a year and one to antero-lateral decompression after three years, had permanent partial paraplegia. One patient, in whom release of a paravertebral abscess after four months and later antero-lateral decompression at twenty-three months were ineffective, ended with a complete permanent paraplegia of grade 3. One patient, rapidly deteriorating at nine months, did not improve after antero-lateral decompression and died five weeks later. One patient treated conservatively for seventeen months deteriorated to grade 3 and died five weeks later. One patient treated conservatively for seventeen months deteriorated to grade 3 and died with sores and toxaemia.

In all, twenty-seven patients were treated conservatively throughout; sixteen costo-transversectomies, thirteen antero-lateral decompressions, three laminectomies and one aspiration of a paravertebral abscess were performed.

The duration of paraplegia before progressive improvement began was of prognostic import, and improvement was often attended by radiological evidence of diminution of a paravertebral abscess. Thus, of the 102 patients with early onset paraplegia, eighty-eight had a paravertebral abscess, and in forty-four of these improvement in the paraplegia closely coincided with absorption of the abscess (Table V). Only progressive improvement is significant. Incomplete paraplegia may vary in grade 1 and grade 2 for as long as two or three years; in such cases the paravertebral abscess persists. Further, even after progressive improvement relapse sometimes occurs. Of the forty-six patients who recovered completely, twenty-three began to improve within three months, twelve between three and six months, ten between six and ten months, and in one exceptional case a patient who had a large paravertebral abscess successfully evacuated by costo-transversectomy five months from the onset did not begin to improve till ten months later, when recovery became complete fairly rapidly.

Of the four patients showing only slight residual paresis, one fluctuated between grade 1 and grade 2 paraplegia for twenty months, one began to improve at two months but later relapsed and the remaining two began to improve at six and ten months. Of the four cases with permanent partial or complete paraplegia improvement began at thirteen months in one, two years in another and not at all in the remaining two.

In brief, it seems that in a case of early onset paraplegia of grade 2 progressive improvement within six months of the onset almost assures complete recovery; onset of progressive improvement at between six and ten months gives the prospect of residua in approaching 20 per cent; if progressive improvement is delayed for more than a year the paraplegia is likely to be permanently partial or complete.

Grade 3—There were eleven patients with early onset of paraplegia of grade 3. Of four with paraplegia of less than one month's duration complete recovery occurred in two treated conservatively, complete slow recovery in one after costo-transversectomy, and there were very slight residua in one after antero-lateral decompression.

Of four patients with paraplegia of from one to three months' duration two recovered completely after conservative measures and antero-lateral decompression respectively, one had slight residual paresis after antero-lateral decompression, and the fourth, showing progressive improvement after antero-lateral decompression, died three months later from cerebral malaria.

One patient showed no response to costo-transversectomy at two months, but began to

improve with antibiotics at six months and recovered completely. One patient with paraplegia of fifteen months' duration, that had been of grade 3 for at least five months, was unaffected by costo-transversectomy and antero-lateral decompression and remained permanently paralysed. The last patient, with paraplegia of two years' duration, that had deteriorated to grade 3 for an undetermined period, regressed to a severe grade 1 with absorption of a small paravertebral abscess on conservative treatment.

Five patients were treated purely conservatively; five antero-lateral decompressions and four costo-transversectomies were performed.

In short, complete recovery appears the rule in early onset paraplegia of grade 3 that has persisted for less than one month, and recovery is probable when paraplegia has persisted for between one and three months. After three months there is a strong probability of significant residual paresis or of permanent paraplegia.

Grade 4—There were ten patients with early onset paraplegia of grade 4. Of four patients with paraplegia of not more than two weeks' duration one died from tuberculous meningitis, two recovered completely after antero-lateral decompression and one with primary disease of the neural arches recovered completely after laminectomy. Of three patients with paraplegia of between two weeks' and two months' duration two recovered completely, one with conservative measures, the other after antero-lateral decompression and the third had residual paresis after costo-transversectomy. Of three patients with paraplegia of four, seven and eight months' duration, two died from tuberculous meningitis (one three months after antero-lateral decompression) and in the third, despite antero-lateral decompression, paraplegia remained complete and permanent. Two patients were treated purely conservatively, and five antero-lateral decompressions, one costo-transversectomy and one laminectomy were performed.

LATE ONSET PARAPLEGIA

There were thirteen cases of late onset paraplegia. Grading was often not so clear-cut as in cases of early onset because paralysis was often incomplete and physical signs were often irregular. Thus in a grade 2 early onset case the supervention of sensory loss, trophic sores, flexor spasms or sphincter disturbance fairly certainly heralds early deterioration to grade 3 or 4. In late onset paraplegia on the other hand it is by no means rare to find retention of a significant degree of voluntary control attended by sensory loss, sphincter disturbance or trophic sores.

Nevertheless, the term late onset paraplegia embraces all cases of paralysis occurring for the first time more than two years from the onset of spinal disease. In fact four distinct types of late onset paraplegia were distinguished in this series, and the type of paraplegia was of more prognostic significance than its grade. The types are as follows.

Type a—In a case of spinal disease of perhaps three or four years' duration, often without extreme destruction or severe deformity, which was pursuing a slowly progressive course towards quiescence, probably under inadequate treatment, there occurs a recrudescence of activity attended by paraplegia. Radiographs show clear evidence of activity in the spinal lesion and usually a paravertebral abscess.

Type b—This is the type in which low-grade, grumbling spinal disease has been present for five to twenty years, and in which treatment has either never been sought or has been inadequate. A gross gibbus makes an angle of less than 90 degrees. Radiographs may show slight evidence of activity, but the disease is never florid and there is no paravertebral abscess, while mottled calcification in the lesion or at the site of an old abscess may indicate past attempts at healing. Some cases are intermediate between types (a) and (b).

Type c—Sometimes paraplegia supervenes upon a gross, long-standing kyphosis, but clinical, radiological and even operative findings show no sign of activity of the disease and suggest that the spinal lesion is in fact completely healed, but usually without bony fusion.

Type d—Just as an early onset paraplegia may rarely persist, after healing of the spinal lesion, in the presence of a persistent paravertebral abscess, so may a late onset paraplegia manifest itself for the first time under similar circumstances.

Significance of the type of paraplegia—Not only is the recognition of the type of paraplegia of more prognostic value than its grade but it may also have an important bearing on treatment. Thus, of five patients with paraplegia of type (a) three (grade 1, grade 1 and grade 2) recovered completely, one (grade 4) recovered incompletely with conservative treatment, and one (grade 1) recovered incompletely after costo-transversectomy and laminectomy.

One patient whose category was intermediate between types (a) and (b), with a grade 2 paraplegia of six weeks' duration that deteriorated to grade 4, recovered completely after antero-lateral decompression.

There were two cases of type (b). One patient, with paraplegia that was originally of grade 3, was treated conservatively and pursued a fluctuating course for two years before improving to the extent that her general condition was good, there were no flexor spasms, she had recovered full control of the sphincters and was learning to walk with calipers and crutches. The second patient, with paraplegia of grade 2, worsened temporarily after antero-lateral decompression, developing incontinence and shin sores, but within a month began to improve and made a partial recovery.

There were three cases of type (c). One patient with paraplegia of grade 2 worsened after antero-lateral decompression and later died; one whose condition had long fluctuated between grades 1 and 2 deteriorated permanently to grade 4 after antero-lateral decompression; and one with grade 3 paraplegia of fifteen months' duration temporarily regressed to grade 2 after antero-lateral decompression, but later relapsed and died with bedsores and toxaemia.

There were two cases of type (d). One patient with paraplegia of grade 3 recovered completely after antero-lateral decompression, and one with grade 1 paraplegia after evacuation of a large psoas abscess.

Of the thirteen patients with late onset paraplegia five were treated purely conservatively, and there were five antero-lateral decompressions, two costo-transversectomies and one laminectomy.

OPERATIVE TREATMENT

Costo-transversectomy—Of the twenty-seven costo-transversectomies, fifteen were carried out primarily on account of a large or tense paravertebral abscess, the radiological shadow of which persisted or was increasing despite adequate conservative measures, and not essentially for the relief of the paraplegia. Such an abscess, failing to regress with quiescence of the bone lesion, constitutes a potential source of recrudescence of disease or of its spread to adjacent vertebral bodies, and should be emptied irrespective of paralysis. In nine instances an abscess was encountered, but none was found in six. In three cases paraplegia was of grade 1 and in twelve of grade 2; all the patients later recovered completely with the exception of one with grade 2 paraplegia who exhibited slight residual paresis.

In twelve cases costo-transversectomy was performed for the relief of paraplegia. A grade 1 paraplegia of nearly three years' duration (abscess) and a paraplegia that had fluctuated between grades 1 and 2 for twenty months (no abscess) both recovered incompletely. Of four patients with grade 2 paraplegia of early onset one (abscess found) recovered completely but slowly, two (no abscess found) suffered permanent partial paraplegia and the fourth (abscess found) had a permanent complete paralysis. Of four patients with grade 3 paraplegias of early onset one (abscess found) was permanently paralysed, one (abscess found) was unimproved after costo-transversectomy but later recovered fully with antibiotics, one (abscess found) made a slow, full recovery, and one (abscess found) was unaffected and later died of cerebral malaria while improving after antero-lateral decompression. One grade 4 paraplegia (abscess) recovered incompletely, and one late onset paraplegia (no abscess) was permanent but partial.

Although in some cases the result of costo-transversectomy may be dramatic, in only three cases did marked improvement follow so closely on costo-transversectomy as to be unequivocally attributable to it, and it must be considered an unreliable operation for the relief of paralysis.

The frequency with which no pus was found need cause no surprise, for a radiological shadow suggestive of an abscess may equally well represent an inflammatory mass or tuberculous granuloma containing little or no pus.

Laminectomy—This was performed in five cases, but in only one, that of a grade 4 paraplegia due to disease confined to the neural arches, was recovery so prompt and complete as to be unmistakably due to the laminectomy.

Antero-lateral decompression—Twenty-nine antero-lateral decompressions were carried out. Thirteen were in cases of grade 2 early onset paraplegia, and seven of these, with paralysis of four, two, two, two, three, five and six months' duration, and two with paralysis of unspecified but brief duration—nine in all—recovered completely. In five of these nine cases the indications for operation were: blistering of the feet with threatened sores; spinal luxation, anaesthesia with trophic sores, and flexor spasms (two cases). In four cases no clear indication is specified. In six of the nine cases marked improvement occurred within seven to ten days of operation and recovery was complete within two months. In one patient slight improvement, and in another moderate improvement, occurred within two weeks, and progressed to complete recovery within six months. In the last case there was marked improvement within a month, but the spinal lesion later relapsed and fluctuating minor signs persisted till healing of the lesion and final full recovery of the paraplegia occurred two years later. Of the four patients with grade 2 paraplegia that failed to recover completely one was unimproved and died suddenly five weeks later. A second was unaffected by antero-lateral decompression, later deteriorated, but recovered with residual weakness after later costo-transversectomy. The third failed to respond to decompression after twenty-three months of paraplegia and remained completely and permanently paralysed. The fourth case, one of long-standing grumbling disease with a grade 1 early onset paraplegia that had latterly deteriorated to grade 2, was really comparable to a type (b) late onset paraplegia. There was no response to antero-lateral decompression and paralysis was permanent but incomplete.

Five operations were performed for grade 3 paraplegia of early onset. One patient with paraplegia of less than one month's duration recovered with slight residua, showing definite improvement within ten days of operation. Of three patients paralysed six weeks, two months and at least three months, the first improved within three weeks and recovered completely, the second with a progressively improving paraplegia died of cerebral malaria three months after operation and the third improved within two weeks and recovered with minor residual weakness. Lastly a paraplegia of fifteen months' duration, of grade 3 for at least five months, remained permanent and complete.

Of five grade 4 early onset paraplegias three (two of about a week's duration, one of two months' duration) recovered fully, improvement occurring in all within seven to fourteen days of operation. A child of one and a half years with grade 4 paraplegia associated with complete spinal luxation died from tuberculous meningitis with the paralysis unrelieved three months after operation; and a further grade 4 paraplegia of eight months' standing remained permanent and complete.

Six antero-lateral decompressions were performed for late onset paraplegia. The first case was that of a boy of seven with Pott's disease for three years, and a grade 2 paraplegia for a month finally deteriorating to grade 4. At the onset of the disease there had been a paravertebral abscess, but when paraplegia supervened there was no gross kyphosis; radiographically there was evidence of activity, but no gross destruction and no paravertebral abscess. At operation the only evidence of activity was one small focus of carious, crumbly bone. Yet improvement occurred within a month and recovery was full. One type (b) case

of two months' duration, which showed at operation much old caseous matter with some more recent, active, oedematous granulations, worsened temporarily after operation with incontinence and shin sores, but improved within a month to recover ultimately with marked residual weakness.

In the type (c) cases there was clinically, radiologically and at operation no sign of active disease, and in all there was very pronounced kyphosis. In one case of paraplegia of grade 2 an anterior bony ridge was removed, but paraplegia deepened and the patient died later with sores, emaciation and toxaemia. In a second case, with paraplegia that had long fluctuated between grades 1 and 2, the dural tube was at operation found flattened and stretched over the angulated anterior wall of the canal: no attempt was made to remove the bony ridge, but the paraplegia deepened permanently to grade 4. In the third case, with grade 3 paraplegia of fifteen months' duration, improvement occurred after operation to the extent that the sores healed, flexor spasms were abolished, the patient was able to sit out in a chair and was measured for calipers, but later he relapsed and died with recurrent sores and toxaemia.

Lastly, a type (d) grade 3 paraplegia recovered completely. At operation a large paravertebral abscess was emptied, but no sign of active bone disease found, and it is probable that a costo-transversectomy would have served as well.

The first antero-lateral decompression in this series was performed in August 1952 and there were no operative or immediate post-operative deaths.

DISCUSSION

No mention has been made of damage to the cord by pathological dislocation. In six cases there was complete luxation, and in six severe subluxation, but, except in one case, this never seemed to be the dominant factor in the causation of an early onset paraplegia. In one case, in which the radiographic appearance suggested that the cord could scarcely have escaped transection, full recovery occurred after antero-lateral decompression.

It seems to be established, as Butler (1935) maintained, that the factors responsible for the development of an early onset paraplegia are the effects of adjacent inflammation, oedema and toxins on the cord, alone or abetted by mechanical compression from inflammatory granulations, debris or pus. As regards the latter, records of cerebro-spinal fluid examination were available in only three cases in this series. In two cases the fluid was clear and colourless, in one slightly turbid with formation of a web coagulum, and in all the pressure rose with jugular compression, the cell count was normal, the protein over 100 milligrams per 100 millilitres and the globulin increased suggesting partial block. Both the toxi-vascular and the mechanical compressive factors regress in most cases as the spinal bony lesion heals, and the paraplegia recovers accordingly. However, should this process be unduly delayed, recovery may be incomplete or the paraplegia remain permanent, and it is evident that if the acute recoverable changes in the cord are not relieved within a certain critical period, further changes ensue which are not reversible.

As Seddon (1935) stated, it is not the grade of paraplegia alone that determines the outcome but a combination of the grade and the duration. Taking the critical period for grade 2 paraplegias as six months, Seddon (1956) suggested that, once voluntary control has been lost, decompression should not be delayed longer than one month. He justified this apparent haste by pointing out that if the primary decompressive measure be a costo-transversectomy, four to eight weeks will elapse before its efficacy is known. If antero-lateral decompression should thereafter prove necessary, with a similar further recovery period, the critical period of six months will be nearing its close. The present series confirms that the critical period for almost certain full recovery is six months, although 80 per cent of the patients recovered completely after six to ten months' paraplegia. Moreover, of the forty-eight patients with grade 2 paraplegia that did recover fully, only twenty-four began to improve within three

months of the onset. Thus, if Seddon's suggestion of operation within a month of onset is adopted, many patients would be subjected to unnecessary operation, and it must not be forgotten that most early onset paraplegias did recover with conservative measures alone even in pre-antibiotic days. Experience in this series suggests that in a case of early onset paraplegia of grade 2 it is expedient to pursue conservative measures for three months; if then there is no improvement and a large paravertebral abscess shadow is failing to decrease or is actually increasing, costo-transversectomy is performed. If pus is not encountered and released antero-lateral decompression is proceeded with at once. Except in the above circumstances conservative measures are persevered with for five months, and, if then there is no evidence of improvement, antero-lateral decompression is carried out. If at any time while under adequate conservative treatment a grade 2 paraplegia deteriorates or threatens to deteriorate to grade 3 or grade 4, antero-lateral decompression is carried out without delay.

Since it seems possible that there may be some advantage in establishing antibiotic control before operation, it is suggested that a patient admitted in a state of grade 3 or 4 paraplegia be given a preliminary trial of conservative treatment, including antibiotics, before operation. In such cases it is considered however that, in the absence of improvement, operative decompression should not be delayed longer than one month in grade 3 paraplegia, and two weeks in grade 4 paraplegia. Frequently the issue is not so clear-cut as suggested above, and careful judgment may have to be exercised in the case of a grade 3 or 4 paraplegia that has supervened on a grade 2 of variable duration, or a paraplegia that has fluctuated between grades 1 and 2 for perhaps two or three years.

Seddon (1935) stated that if there is still some voluntary control the spinal cord is in little danger of irrecoverable damage. Although this is generally true, permanent sequelae may occasionally result, and it is therefore suggested that operation be considered in any grade 1 paraplegia that has persisted for more than twelve months.

It seems, then, that, apart from some variation in opinion as to the time allowed to elapse before its employment, the indications for antero-lateral decompression in early onset paraplegia are fairly well defined and its results eminently satisfactory.

The prognosis in late onset paraplegia is notoriously less favourable, and the problems of treatment greater. At the one extreme the type (a) late onset paraplegia, with clear evidence of spinal activity and often a paravertebral abscess, may be treated on similar lines to the early onset case, the indications for operation being the same and the prognosis almost as favourable. In the type (b) case, however, activity is slow and grumbling, there is no paravertebral abscess, and even at operation one may encounter only minimal active granulation or oedema superimposed on areas of old crumbly, caseous or fibrous quiescent disease. This is in marked contrast to the florid reaction and paravertebral abscess of the early onset case, and it is not at once clear how such minimal activity could be capable of provoking a paraplegia at all. The answer seems to be that under the influence of years of slow, grumbling activity in its vicinity the substance of the cord has undergone some change, for long subclinical, and perhaps ischaemic or degenerative in nature. Thus, if a slight low-grade recrudescence of inflammation in the bony lesion should occur, it acts upon a damaged cord of low reserve.

In the type (c) case there is clinically, radiologically and at operation absolutely no evidence of active disease, so that local inflammation can be excluded as the precipitant cause of the paraplegia. At operation the anterior wall of the spinal canal is acutely angulated and the dura and its contained cord are often flattened and ribboned over this angulation, and it seems probable that stretching and friction of the cord over this bony spur are responsible for the paraplegia. Butler (1935) explained that this is not a primary stretching of an essentially normal cord over a bony spur, but that the cord has shrunk in length at the site of disease in consequence of the change in its substance postulated above—an irreversible change described by Seddon (1956) as a curious atrophy, an interstitial change the result of a chronic ischaemia, but not a gliosis—and so has become secondarily stretched over the angulation.

As regards treatment, the type (c) paraplegia caused by stretching of an irreversibly damaged cord over a bony angulation stands at the opposite extreme to the type (a) paraplegia which is the effect of adjacent inflammation on an essentially recoverable cord. Seddon (1956) emphasised the difficulties of removing such a solid, bony spur, and the dangers of inflicting further damage on the cord in the process. In one case in this series such damage resulted from the approach alone, without any attempt at removal of the bony ridge, possibly from interference with contributory vessels, either epidural or entering through the intervertebral foramina. Of the three patients with type (c) disease in this series one deteriorated and later died after removal of a bony ridge, a second deteriorated to a permanent grade 4 paraplegia after antero-lateral exposure without attempted attack on the spur, and the third, after a modest temporary improvement, relapsed and died some months later—a discouraging record. Although it is possible that more favourable results might be obtained with greater technical expertise, it seems probable that damage to the cord is in the main irreversible, and its residuum of functional tissue exceedingly vulnerable and precarious. Whereas little is to be lost by exploration if a type (c) paraplegia is complete already, I believe that in a case of partial paraplegia the chances of aggravation by operation outweigh those of benefit, and I prefer at present to eschew attempts to remove a bony ridge or to reroute the cord, and confine activities to a fairly extensive posterior fusion: it is hoped that the latter may obviate further increase in the gibbus and limit friction of the cord over the angulation, and so arrest the progress of the paraplegia.

The type (b) case falls intermediate between types (a) and (c), both cord degeneration and adjacent inflammation contributing their quota to the pathogenesis. Although there is some danger that disturbance of the damaged cord may intensify the paraplegia by further restricting its vascular supply, the benefit conferred by local decompression may well outweigh this risk. Some of these patients recover or improve considerably with conservative treatment, which, since the paralysis is often incomplete, may be given a long and generous trial. In these type (b) cases a gross kyphosis with extensive bony destruction often renders final healing protracted and uncertain, and it necessitates immobilisation of prohibitive duration. To facilitate healing and as a safeguard against relapse of the paraplegia, therefore, an extensive posterior fusion should always be undertaken once the paraplegia has responded either to conservative or operative measures.

Inter-body fusion at the time of antero-lateral decompression has not been attempted in this series because of the fear that inadequately fixed grafts might become dislodged and exert pressure on the cord.

In short, in cases of late onset paraplegia, the greater the activity of the bony lesion, the better is the prognosis, and the less are the risks of increased cord damage from antero-lateral decompression.

As regards the type of operation to be employed, laminectomy is desirable only in paraplegia from disease of the neural arches, or when disease of the cervical spine renders antero-lateral decompression inapplicable. Even when a large paravertebral abscess has been successfully evacuated, recovery from paraplegia after costo-transversectomy has proved most uncertain. When recovery does occur it is so late or protracted that it may seem to be coincidental, though undoubtedly a small proportion of prompt and dramatic results are obtained. The use of costo-transversectomy has, therefore, come to be restricted to three types of case: 1) cases of grade 2 early onset paraplegia that has persisted unimproved for three months, with a large paravertebral abscess that has failed to decrease or is increasing despite adequate conservative treatment; 2) the infrequent cases of paraplegia with a paravertebral abscess but without detectable (or with a minimal) bony lesion in the radiographs; 3) paraplegia persisting or arising for the first time with a long-standing residual paravertebral abscess which has persisted unchanged, although the spinal lesion is regressing, quiescent or even healed.

In all other circumstances demanding decompression, the antero-lateral operation is chosen. Unless undue delay or long-standing disease has resulted in irreversible cord change, its results are reliable and usually evident within two weeks of the operation. At operation evacuation of any paravertebral abscess should be ensured. In one case in this series this was not done: improvement did not follow antero-lateral decompression, the paraplegia later deteriorated, and only recovered after costo-transversectomy.

SUMMARY

1. The results of treatment of 115 patients with Pott's paraplegia during a six and a half year period are reviewed.
2. The indications for operative intervention—in particular antero-lateral decompression—according to the duration, grade and type of paraplegia, are discussed.
3. No attempt has been made to justify the treatment of individual patients in the series, some of whom would undoubtedly have benefited from earlier operation; but lessons are pointed and conclusions drawn from the results, good and bad.

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REFERENCES

BUTLER, R. W. (1935): Paraplegia in Pott's Disease, with Special Reference to the Pathology and Etiology. British Journal of Surgery, **22,** 738.
SEDDON, H. J. (1935): Pott's Paraplegia: Prognosis and Treatment. British Journal of Surgery, **22,** 769.
SEDDON, H. J. (1956): Pott's Paraplegia. *In* Modern Trends in Orthopaedics. (Second Series), p. 220. Edited by Sir Harry Platt. London: Butterworth & Co. (Publishers) Ltd.

SPINAL TUBERCULOSIS IN SOUTHERN NIGERIA

With special reference to Ambulant Treatment of Thoraco-lumbar Disease

PETER G. KONSTAM and SHEILA T. KONSTAM, IBADAN, NIGERIA

From University College Hospital, Ibadan

Thoraco-lumbar tuberculous spondylitis is prevalent in Southern Nigeria. Ibadan, a town of half a million people, has hitherto been served by two small hospitals with a totally inadequate number of beds. It was clear that no accommodation could be afforded for patients with the prospect of a prolonged stay if beds were to be kept available for still more urgent work. It was therefore decided to attempt treatment of spinal tuberculosis by an ambulant out-patient régime except when there was evident or suspected involvement of the spinal cord.

In the first part of this paper the general pattern of tuberculous spondylitis as seen in 140 South Nigerians is outlined briefly. In the second part the method and results of out-patient treatment of thoraco-lumbar disease are described.

CHARACTERISTICS OF THE DISEASE

Bacteriology—Though no bacteriological data are as yet available it is a fact that, after weaning, no fresh milk is consumed in Ibadan because there is still no trypanosoma-resistant breed of

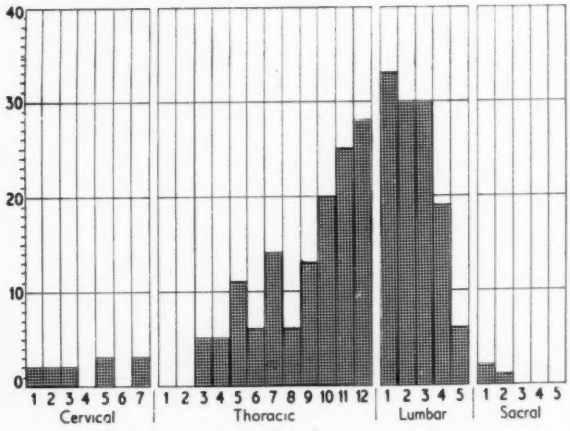

FIG. 1

Histogram showing the incidence of vertebral tuberculosis at various levels.

cattle that can be kept on a sound commercial basis in these regions. There appears to be no clear source of bovine tuberculous infection and, in fact, classical calcifying tuberculous mesenteric adenitis (tabes mesenterica) is hardly ever seen. Inhalation is presumably the mechanism of infection.

Tuberculin sensitivity—The " Heaf test " was positive in all cases.

The lungs—Sixty-two of the patients (40 per cent) had radiological evidence of pulmonary tuberculosis. In thirty, this was a hilar adenopathy. In thirty-two there were radiological signs of parenchymal disease.

Associated non-pulmonary tuberculous lesions—Four patients had phlyctenular conjunctivitis, one " caries sicca " of the shoulder, one " caries sicca " of the hip and one tuberculous abdominal glands.

Complications—Sinuses or psoas abscess were seen in eight patients. None appeared during treatment. Nine patients had involvement of the spinal cord, which developed under treatment in two. One child died of tetanus thirteen months after the beginning of treatment. The source may have been a sore under the plaster jacket. Patients ought to be immunised before jackets are applied because tetanus is common in Southern Nigeria.

Localisation—Altogether 163 patients with tuberculosis of bone and joint were seen since 1952. Of these 140 had spondylitis. In the remaining twenty-three the disease was localised as follows: knee nine, hip five, shoulder three, hand three, elbow two and ankle one. The preponderance of spinal lesions is clearly seen. Of the 140 patients with spinal disease the cervical spine was involved in ten and the sacral vertebrae in three.

Dobson (1951), in a large series, found the mid-thoracic region affected in 45 per cent of cases, the lower thoracic region in 15 per cent and the lumbar region in 31 per cent. A functional grouping of the lower thoracic segments with the lumbar segments may be preferable to a descriptive anatomical subdivision of the spine. The region T.10–L.4, being the mobile segment, might be considered as one functional unit. Our histograph (Fig. 1) shows this unit predominantly affected in our patients. The frequency with which L.1, 2 and 3 are affected indicates a deviation from the European pattern.

Age—The age incidence is shown in Table I.

TABLE I

AGE INCIDENCE (140 PATIENTS)

Age in years	Number of patients
0–4	44
5–9	52
10–14	20
15–25	4
25 and above	20

Nigerians commonly carry heavy loads on their heads. It has been suggested that this may predispose their spines to disease. The large number less than five years old makes this supposition unlikely.

Sex—There were eighty-six males and fifty-four females.

Bridge formation—An unusual feature in eight of our older patients was the formation of bridges in the anterior and lateral spinal ligaments. These were not due to calcification of a paravertebral abscess: true bone had formed. The sacro-iliac joints were normal in all.

In three patients bridges formed during out-patient treatment.

Case 1—In 1954 a woman aged twenty-seven was found to have caries of T.10, 11, 12 and L.1 vertebrae, with pulmonary tuberculosis. A year and a half later there was an anterior bridge from L.2 to fused T.12 and L.1; T.10 and 11 appeared healed (Fig. 2).

Case 2—In 1956 a man aged twenty-two developed caries with collapse of T.10 and 11 vertebrae. A year and a half later a bridge was forming between L.1 and L.2.

Case 3—In 1956 a boy aged fifteen was seen with caries of T.12 and L.1 vertebrae. The diseased L.4 vertebra was joined to L.3 and nearly to L.2; there was " caries sicca " of the left shoulder. A year later there was healing of T. 12 and L.1 with a new bridge; L.2 and L.3 vertebrae were solidly united (Fig. 3).

FIG. 2
Case 1—Marked anterior bridging eighteen months after onset.

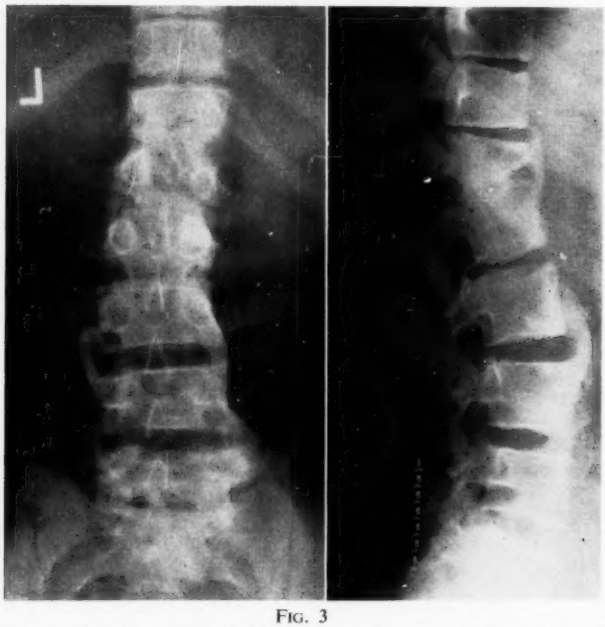

FIG. 3
Case 3—Extensive anterior bridging a year after onset of tuberculosis of T.12 and L.1 vertebrae.

In one patient solid bridge formation was present at the time of first attendance. A similar state was present in one further patient but bridge formation continued under out-patient treatment.

In contrast, in three patients referred to our clinic bridge formation was present without evidence of spinal caries. Thus in a man aged fifty there was bridge formation in the thoraco-lumbar spine, and a sinus over the sternum.

In the two patients in whom bridges were present at the time of the first attendance the caries occurred above the level of the bridge, possibly from abnormal stresses. In some cases of bridging the affected vertebrae showed no signs of past or present caries. In three patients (1, 2 and 3) bridge formation, although at a distance from the disease, appeared as part of the healing process, and may be taken as an expression of a strong tendency to reactive bone formation which will be referred to later. Further investigation is necessary.

FIG. 4
Some of the patients.

OUT-PATIENT TREATMENT

We excluded ten patients with cervical caries as we felt that these should be treated by orthodox methods. Only thoraco-lumbar and sacral disease was considered suitable. Eight patients belonging to this group had burnt-out lesions. There remained 122 patients who were accepted for ambulant treatment.

The first group of patients comprised six children who attended first in 1952. They were given rest in hospital for one month during which they received streptomycin and para-amino-salicylic acid and were put into plaster-of-Paris jackets. Drug treatment was discontinued thereafter and the children were discharged home. They were seen at three-monthly intervals for assessment and reapplication of plaster.

Under this régime the disease process took a favourable turn in three patients, but in the other three it continued to advance. In all but one there was spread of the disease after treatment began, one further vertebra being affected in three and two further vertebrae in two patients.

Wilkinson (1954) stated that direct observation of tuberculous tissue treated with streptomycin showed little improvement in established osseous lesions. He concluded that streptomycin and iso-nicotinic acid hydrazide cured early and mainly synovial lesions but not late ones. Our patients do not attend for early lesions of any sort. As it appeared that

FIG. 5

FIG. 6

Case 6—Tuberculous spondylitis at T.9–10 in a girl of four, treated as an out-patient. Figure 5—Initial radiograph. Figure 6—Nine months later. Evidence of healing.

our régime was not likely to produce the desired results we began to institute prolonged drug treatment of the simplest kind in our out-patient department along with plaster immobilisation, eliminating streptomycin injections and the initial stay in hospital (Fig. 4). Drugs were given as follows: *para-amino-salicylic acid*—Children under two years received four grammes daily; those aged two to four years, six grammes daily; aged four to ten years, six to nine grammes daily. *Isoniazid*—Five milligrams per pound of body weight per day.

Of the 116 patients accepted for this new régime, seventeen are still in the early phase of treatment. Of the remaining ninety-nine, thirty-eight abandoned treatment, and in nine it was unsuccessful. Of these, spread during treatment occurred in three, two patients developed paraplegia, and in one the disease remained active after a year's treatment. There

were three deaths, one from tuberculous broncho-pneumonia two days after the first attendance, and two from unknown causes.

Fifty-two patients did well or are dong well. The following are examples of successful ambulatory out-patient management of lumbo-thoracic tuberculous spondylitis.

Case 4—In 1956 a boy aged seven years was found to have T.5 and T.12 vertebrae separately affected by tuberculosis. A year later T.5 vertebra was collapsed and solidifying.
Case 5—In 1956 a girl aged five years was seen with caries of T.6–8 vertebrae and a paravertebral abscess. A year later there was evidence of healing, and the paravertebral abscess was shrinking.

FIG. 7

FIG. 8

Case 10—Girl aged two with tuberculosis at L.4–5. Figure 7—Initial radiograph. Figure 8—Sixteen months later.

Case 6—In 1956 a girl aged four years developed caries of T.9 and 10 vertebrae (Fig. 5). Five months later the disease process was continuing, with vertebral collapse. After a further four months there was merging of the vertebrae with healing (Fig. 6).
Case 7—In 1955 a girl aged two years attended with severe caries, with collapse of T.10, 11 and 12 vertebral bodies. Four months later T.11 vertebral body had disappeared, and the disease was quiescent.
Case 8—In 1955 a boy aged two years attended with caries of T.12 and L.1 vertebrae. A year and a half later there was fusion of the affected wedged vertebrae with healing.
Case 9—In 1955 a girl aged two years developed caries of L.2 and 3 vertebrae. Seven months later there was merging of the affected vertebrae, with sound union.
Case 10—In 1955 a girl aged two years was seen with destruction of L.4 and 5 vertebrae (Fig. 7). A year and four months later there was fusion of the affected vertebrae and sound healing (Fig. 8).

DISCUSSION

The ambulant treatment of patients suffering from tuberculous spondylitis was forced upon us by circumstances. Healing has often been rapid, and there have been few reverses.

Drug treatment appears to be more important than plaster fixation: plaster jackets are not well looked after by some of our patients and, especially during the rainy season, become soft. The main purpose of the jacket is relief of pain—for which it may be dramatically effective—and prevention of gross deformity.

It may be that West Africans have a special tendency to reactive bone formation (ununited fractures are uncommon among them) but it is also possible that, irrespective of race, maintaining the upright posture may be less harmful in Pott's disease than is commonly supposed. Maintenance of weight bearing, an active circulation and a good morale may prove equal or even superior to the long-established opposite. In any case the method adopted at Ibadan may be of use in those underdeveloped countries where prolonged retention in hospital cannot be attempted.

SUMMARY

1. In Nigeria, among the sites affected by tuberculosis of bones and joints there is especial frequency in the lumbo-thoracic junction from the tenth thoracic to the fourth lumbar vertebrae.

2. Outstanding in the pattern of the disease in adults is a tendency in certain patients to ossification of the spinal ligaments.

3. An ambulatory out-patient régime is described. So far it has promised well.

Thanks are due to Dr J. H. Middlemiss and Dr P. Cockshott for radiological advice, and to Dr A. Tompkins and Dr R. G. Hendrickse for their collaboration.

REFERENCES

DOBSON, J. (1951): Tuberculosis of the Spine. An Analysis of the Results of Conservative Treatment and of the Factors Influencing the Prognosis. Journal of Bone and Joint Surgery, **33–B,** 517.

WILKINSON, M. C. (1954): Chemotherapy of Tuberculosis of Bones and Joints. Journal of Bone and Joint Surgery, **36–B,** 23.

CONGENITAL VERTICAL TALUS

G. C. LLOYD-ROBERTS and A. J. SPENCE, LONDON, ENGLAND

From the Orthopaedic Department, the Hospital for Sick Children, Great Ormond Street, London, and the Institute of Orthopaedics and Royal National Orthopaedic Hospital, London and Stanmore

Congenital vertical talus is an uncommon but well known cause of severe flat foot in children. It is characterised by distinct clinical and radiological features which are, however, sometimes simulated in cases of flat foot from other causes, known or unknown. Our interest was aroused when we found difficulty in distinguishing between this truly congenital abnormality and severe flat foot with talo-navicular subluxation. It seemed desirable therefore to review our cases critically and to establish criteria upon which we could differentiate the one from the other, preparatory to a study of the natural history of the anomaly and the methods of treatment available. In this communication we are concerned almost exclusively with the diagnosis and differential diagnosis of congenital vertical talus—other observations will be incidental.

Although there are numerous continental papers on congenital vertical talus there are few in English. In most, emphasis has been on treatment rather than on the clinical aspects. More recently however, in discussing treatment, Hark (1950) and Osmond-Clarke (1956) have mentioned some of the clinical features. The literature up to 1939 was well reviewed by Lamy and Weissman (1939).

CLINICAL MATERIAL

We have classified thirty-two feet in twenty-two patients as examples of congenital vertical talus. We offer no apology for reporting so small a number, for we find ourselves in company with others who have been critical in accepting this diagnosis. Osmond-Clarke (1956) reported an incidence of one in 121 in a special clinic for congenital foot deformities. Our patients have been traced by reference to the diagnostic indices of the Hospital for Sick Children (seventeen) and the Royal National Orthopaedic Hospital (five)—covering eighteen and twenty-seven years respectively. Their ages are between infancy and fifteen. Few of the severe flat feet seen at the Hospital for Sick Children are radiographed, and it is probable that some cases of vertical talus were classified under other diagnoses. The facts that have led to our conclusions about diagnostic criteria and differential diagnosis have come not only from these patients but also from a study of a far greater number of severe flat feet of varying causes.

THE CLINICAL AND RADIOLOGICAL FEATURES OF CONGENITAL VERTICAL TALUS

The deformity, as its name implies, is present at birth and is usually noticed within the first few weeks. From the first there is a prominence in the sole from which the heel and forefoot rise in a gentle curve (Figs. 1 and 2). The forefoot is dorsiflexed; so a diagnosis of congenital talipes calcaneus is commonly made if the associated equinus deformity of the heel is overlooked (Figs. 1 and 3). Deep creases may be present in front of the ankle (Fig. 3) but eversion of the heel and abduction of the forefoot are not conspicuous in infancy. From the first the deformity is fixed and does not yield to forceful manual pressure. The radiograph is characteristic at a very early age (Fig. 1) for it shows not only the curved outline of the soft tissue of the sole and dorsiflexion of the forefoot, but also a vertical talus and a calcaneum that points into equinus.

When walking begins—this is not apparently delayed—the foot is obviously flat. It is also rigid, so that the foot looks almost the same whether weight bearing or not. The heel

FIG. 1

FIG. 2

Figure 1—Congenital vertical talus in an infant of four weeks. Note the perpendicular talus and the equinus calcaneum. The soft-tissue outline of the sole and dorsiflexion of the forefoot are seen. (A normal foot is shown in Figure 2.)

becomes everted and the forefoot becomes abducted on the medially displaced and now readily palpable talus. The heel remains in equinus and the posterior part of the calcaneum fails to touch the ground on standing (Fig. 4). Development of the tarsal bones now permits recognition of some of the specific radiological features (Figs. 5 to 7). The calcaneum is narrower at its anterior end and becomes somewhat beak-shaped. The talus, lying almost in line with the tibial shaft, becomes narrow at the waist and resembles an hour-glass. When the navicular bone shows it appears to be dislocated from the head of the talus and lies in contact with the narrowed body of the bone (Fig. 6). The dislocated navicular bone carries with it the remaining tarsal and metatarsal bones, so that there is dorsal displacement of the foot at the mid-tarsal joint. Elevation of the first metatarsal bone may cause plantar flexion and rigidity of the first metatarso-phalangeal joint (Fig. 4). Osteochondritis of the navicular (Fig. 6), with the changes typical of Köhler's disease, sometimes occurs. The navicular finally becomes wedge-shaped towards the sole (Fig. 7). In the antero-posterior radiograph the talus projects widely to the medial side of the calcaneum.

FIG. 3

Appearance of foot at four months. Note the heel in equinus and the forefoot in dorsiflexion. A deep crease is seen in front of the ankle.

In later childhood the symptoms are an awkward and clumsy gait and distortion of the shoes: fixed valgus deformity of the heel causes rapid distortion of the uppers, and the heels are constantly in need of repair on their inner sides. There may also be symptoms from associated abnormalities, which were present in more than half the patients reported here. Pain appeared for the first time at the age of twelve in one patient and at thirteen in another.

In summary, diagnosis rests upon the shape of the foot, coupled with its rigidity and a characteristic radiograph. If doubt arises on looking at the radiograph the cause of the flat foot is likely to be other than congenital vertical talus.

CLINICAL TYPES OF CONGENITAL VERTICAL TALUS

The patients that we have examined fall into four groups, according to the presence or absence of associated defects, and their nature: 1) with arthrogryposis (ten patients); 2) without associated defect—that is, the vertical talus is an isolated congenital defect (nine

FIG. 4

Congenital vertical talus in a child of six years showing the characteristic boat-shaped foot and the point of the heel off the ground. Note the plantarflexion of the big toe compensating for the elevated first metatarsal head.

FIG. 5

Aged three years—illustrating the position and shape of the talus, the equinus calcaneum and the dorsiflexed forefoot. The outline of the sole can be seen in this standing radiograph.

patients); 3) with spina bifida (two patients); 4) with neurofibromatosis (one patient). There is no difference between these groups clinically or radiologically, so far as the feet are concerned.

Arthrogryposis is so often present that it is tempting to speculate on its place in the etiology of those cases that apparently exist as single abnormalities or with spina bifida.

Both types have in common the rigidity and resistance to treatment that characterise manifest arthrogryposis. It is unfortunately not possible to establish the truth or otherwise of the hypothesis that arthrogryposis plays a part.

The deformities associated with arthrogryposis were protean and included five dislocated

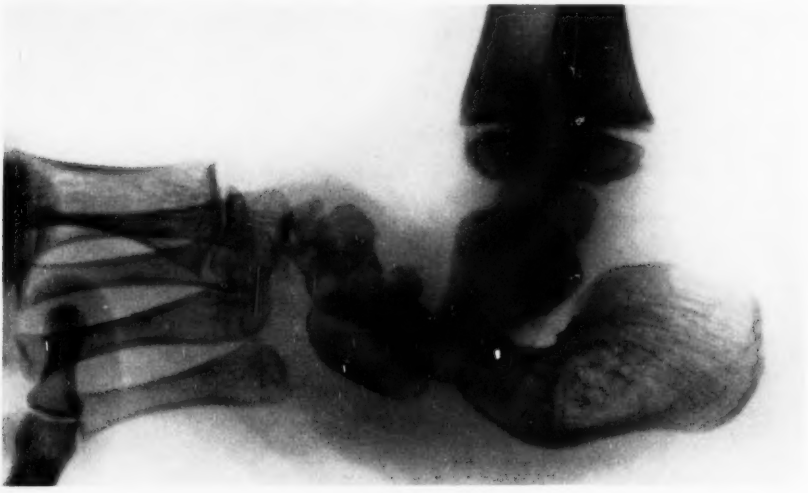

FIG. 6

Aged six years—illustrating the features shown in Figure 5 together with the navicular dislocation and epiphysitis.

FIG. 7

Aged twelve years—the bones are approaching maturity and retain the features of early childhood in addition to the wedging of the navicular and the beak-shaped calcaneum.

hips. Denis Browne (personal communication) stresses the association between valgus feet of any type in infancy and congenital dislocation of the hip. This observation is supported by our findings in this arthrogrypotic group.

Spina bifida was in one case manifest (with a meningocele) and in the other occult.

In patients with meningoceles the deformity may be more common than it appears here, because, being often relatively unimportant, it may be overlooked. Figure 1 illustrates the foot of an infant in whom spina bifida occulta was found. The implication is that the deformity is not simply an extreme example of paralytic flat foot; that deformity appears later, has a

TABLE I

INCIDENCE OF ASSOCIATED DEFECTS IN TWENTY-TWO PATIENTS WITH CONGENITAL VERTICAL TALUS

Clinical type	Patients	Feet
In association with arthrogryposis	10	16
No associated abnormality	9	13
In association with spina bifida	2	2
In association with neurofibromatosis	1	1
Total	22	32

constant pattern of paralysis not present in these cases, is progressive, and lacks distinctive radiological features.

The one example of vertical talus occurring in association with neurofibromatosis is a curiosity not mentioned by McCarroll (1950) in his review of skeletal abnormalities associated with this disease.

FIG. 8

Severe idiopathic flat foot in a child of four. The heel touches the ground.

DIFFERENTIAL DIAGNOSIS

Idiopathic flat foot—The deformity on standing may approach in severity that seen in congenital vertical talus, but the whole lower surface of the heel remains on the ground (Fig. 8). At rest the foot does not appear deformed, and the valgus heel, which is not in equinus, can be placed readily in a neutral position. In early childhood the head of the talus can be pushed up from

the sole and, like the valgus heel, it offers no resistance to passive correction until early adolescence, when inversion at the subtalar joint may become restricted. There may, however, be early limitation of dorsiflexion, detectable only when the heel is held in the neutral position,

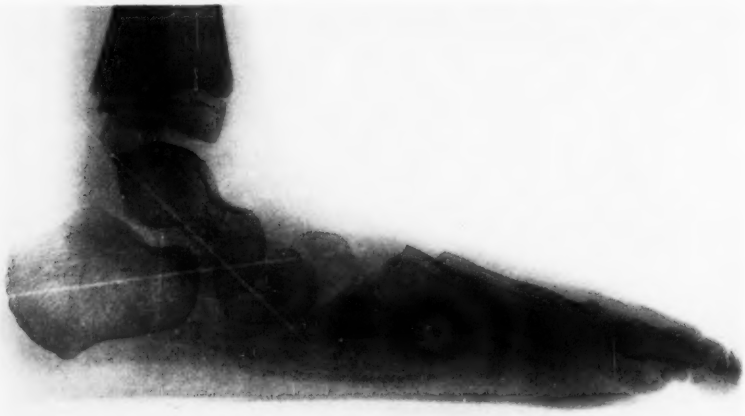

FIG. 9

Severe flat foot with talo-navicular subluxation in a child of five years. The talus inclines towards the perpendicular but there are no other abnormalities in the standing radiograph.

FIG. 10

The same patient as in Figure 9, radiographed on the same day, but with the film badly centred and the patient not bearing weight. Although the talus is vertical there are no other abnormalities.

and caused by shortening of the calcaneal tendon, either primary or secondary (Harris and Beath 1948). The prominence of the head of the talus in a mobile flat foot does not precede weight bearing, whereas the prominence with congenital vertical talus is present at birth, fixed from the start, and no worse on weight bearing.

Any doubts can be removed by radiography with the patient standing (Jack 1953). In idiopathic flat foot the talus often becomes displaced towards the sole, but there is never complete talo-navicular dislocation, nor does the talus approach the perpendicular. The calcaneum may be horizontal if the posterior calf muscles are short, but it is never in equinus (Fig. 9). Secondary changes in the body of the talus, navicular and calcaneum do not occur,

FIG. 11
Severe flat foot in cerebral palsy.

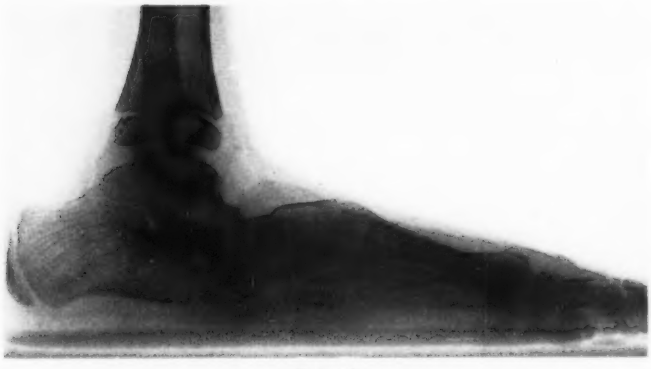

FIG. 12
Radiograph of the foot illustrated in Figure 11. Cerebral palsy in patient aged twelve years. The talus is steep but has neither the characteristic shape nor the other signs shown in Figure 7. The calcaneum is horizontal and the heel touches the ground.

and the forefoot is not dorsiflexed. Mistakes may arise if the radiograph is incorrectly centred, as in Figure 10. On this evidence congenital vertical talus was diagnosed in spite of the lack of changes, such as talo-navicular dislocation. Figure 9 shows the same foot radiographed while the patient was standing.

Paralytic flat foot—This may be caused by anterior poliomyelitis or by spina bifida. Clinical

examination should detect the weakness of the invertors, but even if this is difficult to discover in infancy the mobility of the foot should prevent confusion with congenital vertical talus. When rigidity in eversion develops later it is mainly the shortening of the tendo calcaneus that tethers the calcaneum in valgus, rather than a fixed eversion of the subtalar joint. The radiograph resembles that of severe idiopathic flat foot.

Flat foot in cerebral palsy—Flat foot is a common accompaniment of cerebral palsy and a minor disability in a severely handicapped patient. Nevertheless in very young children with mild cerebral palsy it is an important sign that may be helpful in diagnosis. Confusion with congenital vertical talus may arise because the equino-valgus deformity may become fixed at an early age, but the radiograph again resembles that of idiopathic flat foot. The calcaneum may be horizontal but the heel does not leave the ground (Figs. 11 and 12).

FIG. 13

Uncorrected club foot in patient aged three years. The calcaneum is in equinus. Compare with Figure 5.

Spurious correction of a club foot—When treatment of talipes equino-varus results in spurious correction of equinus at the midtarsal joint the head of the talus becomes prominent and fixed in the sole. In the radiograph the talus and calcaneum are seen to remain in equinus, and the picture may simulate congenital vertical talus. When inversion of the heel has been satisfactorily corrected the resemblance is even greater (Fig. 13). The distinction is likely to be made on the history, and may be supported by the radiograph which shows this iatrogenic vertical talus to differ from the congenital by its broad top and the absence of narrowing of the body.

Talipes calcaneo-valgus—This mistaken diagnosis was made in six infants. It is probable that dorsiflexion of the forefoot distracted the examiner's attention from the equinus deformity of the heel.

TREATMENT

Fifteen out of twenty-two patients had been treated. We have summarised in Table II the non-operative methods that were used, together with the number of patients treated by each technique; more than one method was used in most patients.

Clearly these methods are too diverse for individual assessment; but they have in common an unsatisfactory outcome, because the deformity was neither corrected nor significantly improved in any instance even when treatment started within the first few weeks of life.

Operative methods—Tenotomy of the tibialis anterior was performed three times without apparent benefit. Wedge tarsectomy failed to correct the deformity in one child of seven and is to be repeated. Dunn's arthrodesis was carried out twice for pain and deformity in adolescents both aged fourteen, and after a short period of observation the outcome in these is promising.

TABLE II

METHODS OF NON-OPERATIVE TREATMENT USED IN TWENTY-TWO PATIENTS (TWENTY-EIGHT FEET)

Denis Browne hobble splint, reversed	6
Manipulation followed by plaster .	6
Iron and T-strap	4
Shoe wedges and stiffening . .	5
Corrective night splints . . .	7

Treatment was not attempted in seven patients, because of mental deficiency or some other dominating disability and the pessimism that followed analysis of our results.

SUMMARY AND CONCLUSIONS

1. The clinical and radiological features of thirty-two feet with congenital vertical talus are described and subdivided into groups determined by the presence or absence of associated abnormalities.

2. The differential diagnosis of congenital vertical talus from flat foot, talipes calcaneus and uncorrected club foot is discussed.

3. No benefit came from either non-operative treatment or tenotomy of the tibialis anterior.

It gives us pleasure to thank Mr Denis Browne for his interest and for his permission to review patients under his care at the Hospital for Sick Children, Mr K. I. Nissen for permission to include two of his patients, Mr A. T. Fripp and Mr David Trevor for allowing us to review one patient each and the latter for Figure 6. We are indebted to Mr J. H. Shelswell for providing us with an early radiograph of one of our cases (Fig. 1). Mr H. Jackson Burrows has given us valuable help in the preparation of this report. We would like to thank Mr Derek Martin, who was responsible for the illustrations, and the Medical Record Department of the Royal National Orthopaedic Hospital.

REFERENCES

BROWNE, D. (1956): Personal communication.

HARK, F. W. (1950): Rocker-foot due to Congenital Subluxation of the Talus. Journal of Bone and Joint Surgery, **32–A**, 344.

HARRIS, R. I., and BEATH, T. (1948): Hypermobile Flat-foot with Short Tendo Achillis. Journal of Bone and Joint Surgery, **30–A**, 116.

JACK, E. A. (1953): Naviculo-cuneiform Fusion in the Treatment of Flat Foot. Journal of Bone and Joint Surgery, **35–B**, 75.

LAMY, L., and WEISSMAN, L. (1939): Congenital Convex Pes Valgus. Journal of Bone and Joint Surgery, **21**, 79.

MCCARROLL, H. R. (1950): Clinical Manifestations of Congenital Neurofibromatosis. Journal of Bone and Joint Surgery, **32–A**, 601.

OSMOND-CLARKE, H. (1956): Congenital Vertical Talus. Journal of Bone and Joint Surgery, **38–B**, 334.

THE SPEED OF HEALING AFTER FRACTURE OF THE TIBIAL SHAFT *

H. ELLIS, OXFORD, ENGLAND

From the Royal Infirmary, Sheffield

Although there is a considerable literature on possible factors concerned in the speed of fracture healing, surprisingly few controlled clinical studies have been carried out to assess their importance. Such investigations are only valid when fractures are compared which differ solely in the particular variable under consideration; any significant difference in the speed of union between two such groups can then be attributed entirely to this factor.

The present study attempts to investigate the effects of certain factors—particularly severity of injury, skeletal traction and distraction—on fracture healing, using this technique in a consecutive series of tibial shaft fractures.

CLINICAL MATERIAL, CLASSIFICATION AND OBSERVATIONS

During the six-year period 1948 to 1953, 576 freshly sustained fractures of the tibial shaft were admitted to the Royal Infirmary, Sheffield (567 patients). Of these fractures, 535 were available for the study of primary fracture healing in normal bone. Nine others, to be considered separately, were treated by bone grafting for delayed union or non-union. The remaining thirty-two fractures comprised those necessitating immediate amputation, pathological fractures and fatalities from overwhelming injuries.

A preliminary survey revealed invariable and rapid healing of tibial fractures in patients under the age of sixteen years; in no instance in this group was immobilisation required for more than twelve weeks. The patients have, therefore, been divided by this arbitrary figure of 16 into children (192 fractures) and adults (343 fractures), the main study being concentrated on the adult group.

TABLE I

SPEED OF UNION AND INCIDENCE OF DELAYED UNION IN FRACTURES CLASSIFIED ACCORDING TO SEVERITY OF INJURY

Severity of injury	Number of fractures	Average time of union (*weeks*)	Cases of delayed union (more than 20 weeks)
Minor	98	10	2† (2%)
Moderate	200	15	22 (11%)
Major	45	23	27 (60%)
Total	343		

† This includes one fracture for which prolonged immobilisation was required because of an associated major fracture of the femur in the same limb.

The fractures in adults fall into three groups according to the treatment employed: 1) 222 fractures were treated by immobilisation only; 2) 112 longitudinally unstable fractures were treated by plaster immobilisation supplemented by skeletal traction through the calcaneum; 3) nine unstable fractures were treated by fixation with one or more screws together with plaster immobilisation.

* Being part of the substance of a thesis accepted for the degree of M.Ch.(Oxon).

The severity of the initial injury—Three variables were used in this study to assess severity of tissue trauma—namely, the degree of fracture displacement, the extent of the compound wound and the severity of comminution.

In a detailed investigation of this series of fractures (Ellis 1956) it was found that minor degrees of compound wounding or of comminution had no demonstrable effect on the speed of union; the rate of healing of such fractures did not differ from that of otherwise similar, but closed and non-comminuted fractures. The three factors which did delay healing in a statistically significant manner were complete displacement, severe compound wounding and severe comminution. In effect, the average time required for union of the most severe fractures was more than twice that of the relatively minor injuries, as will be detailed later.

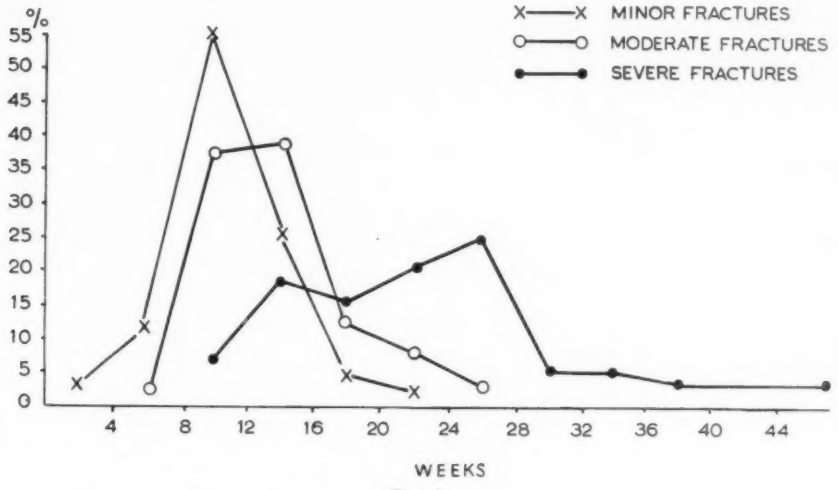

FIG. 1

Histogram demonstrating the rates of union of tibial shaft fractures of minor, moderate and major severity. The percentage of fractures that were united at the end of each four-week period is shown for each group.

These findings have been combined to enunciate a simple classification of fractures into three groups of severity, defined as follows: 1) *Minor severity*—fractures with undisplaced or angulated fragments, whether or not complicated by minor degrees of comminution or compound wounding. 2) *Moderate severity*—fractures with completely displaced fragments, whether or not complicated by minor degrees of comminution or compound wounding. 3) *Major severity*—fractures complicated by major comminution or a major compound wound. (In practice these were all found to be completely displaced.)

Table I shows the average time of union* and the incidence of delayed union† in these three grades of severity of injury, the average times being ten, fifteen and twenty-three weeks respectively and incidence of delayed union 2 per cent, 11 per cent and 60 per cent respectively ($P = < 0.01$). These differences are illustrated by the histogram (Fig. 1) which demonstrates

* Clinical union is said to have occurred when pressure over the fracture and "springing" the bone failed to elicit pain. Naturally, this measurement allows wide variations. However, all the patients were treated under the supervison of one consultant and a small number of registrars, so that a relatively constant level of assessment may be considered to have existed.

† Delayed union is said to have taken place when clinical union of the fracture occurred after more than twenty weeks' immobilisation.

the certain and rapid healing of minor fractures, the increasing scatter and delay in moderate fractures, and the marked shift to the right of the chart in fractures of major severity.

Now let us consider other factors which may affect the speed of fracture union, using this classification as a means of providing groups of fractures of comparable severity of tissue trauma.

Infection occurred in five of the 105 compound fractures which proceeded to clinical union. The numbers are too small to be significant, but bone healing took place within the same limits of time as non-infected fractures of comparable severity treated by similar methods. Certainly, infection alone could not account for the delays in healing of major compound fractures in the present study.

Distraction (defined as a gap of one-sixteenth of an inch or more between the fragments, measured on the radiograph after reduction) was present in eighteen of the 112 fractures treated by skeletal traction (16 per cent.) Distracted fractures took, on the average, five

TABLE II

COMPARISON BETWEEN RATES OF UNION OF DISTRACTED AND NON-DISTRACTED FRACTURES OF COMPARABLE SEVERITY TREATED BY SKELETAL TRACTION

Severity of injury	Number of fractures	Average time of union (*weeks*)	Delayed union (more than 20 weeks)
Moderate			
Distracted	13	19	6 (46%)
Not distracted	74	14	6 (8%)
Major			
Distracted	5	30	5 (100%)
Not distracted	19	25	11 (58%)
Total	111*		

* One fracture excluded because no immediate post-reduction radiographs are available.

weeks longer to unite than comparable fractures treated by traction without distraction ($P = <0{\cdot}01$). The risk of delayed union was also considerably increased (Table II).

Traction—To assess whether skeletal traction, in which distraction has been avoided, affects the speed of union, I compared fractures treated by plaster immobilisation alone with cases of comparable severity treated by calcaneal traction, excluding cases with distraction (Table III). There was no significant difference between comparable fractures of moderate severity treated by these methods ($P = 0{\cdot}35$). In major fractures, although the incidence of delayed union was comparable, there was a lag (five weeks) in the average rate of healing in the cases treated by traction ($P = <0{\cdot}01$).†

Age of the patient—It has already been noted that tibial fractures in patients under the age of sixteen united with uniform rapidity. Above this age, no statistical difference existed between the various age groups, providing fractures of comparable severity were studied.

Site of fracture—With one exception, fractures of comparable severity at various sites of the

† One should note that this difference was due to two fractures with extremely severe tissue damage included in the traction group. Both were initially borderline cases for immediate amputation, both were subject to marked delay in healing (fifty and fifty-six weeks) and both were without equivalent in the cases treated by immobilisation only. If these two cases are allowed for, there was an average lag of two weeks in the major fractures treated by traction compared with similar fractures treated by immobilisation, there being no statistical difference between the two groups ($P = 0{\cdot}65$).

tibial shaft showed no significant difference in speed of union. This exception occurred in fractures immediately below the tibial condyles; only fractures of minor or moderate severity occurred at this site, but these united without a single example of delayed healing.

Fractures with delayed or non-union treated by bone grafting—Of the five fractures with established non-union in the series, two had been treated by traction and three by immobilisation alone. Of four fractures with delayed union treated by bone grafting, two had been subjected to traction (one revealing radiological evidence of distraction) and two to immobilisation alone. Of these nine fractures, no less than eight were injuries of major severity. The remaining one, a moderate fracture with established non-union, was found at operation to have muscle interposed between the fragments.

TABLE III

COMPARISON BETWEEN RATES OF UNION IN FRACTURES OF COMPARABLE SEVERITY TREATED BY IMMOBILISATION ONLY AND IMMOBILISATION SUPPLEMENTED BY SKELETAL TRACTION
(Distracted cases excluded)

Severity of injury	Method of treatment	Number of fractures	Average speed of union (*weeks*)	Delayed union (more than 20 weeks)
Minor	Immobilisation only	98	10	2 (2%)
	Traction	—	—	—
Moderate	Immobilisation only	104	15	12 (11%)
	Traction	74	14	6 (8%)
Major	Immobilisation only	20	20	11 (52%)
	Traction	19	25	11 (58%)
Total		315*		

* Excludes eighteen distracted fractures.

DISCUSSION

The severity of injury—Several authors have suggested that the severity of injury is an important determinant of the speed of fracture healing (Carpenter, Dobbie and Siewers 1952, Griffiths 1942, Starr 1947, Wilson 1942). This suggestion has been confirmed and extended in the present investigation.

Although minor degrees of comminution or compound wounding had no effect on the speed of fracture healing in the cases that I studied, these complications become of considerable importance when they indicate, by their severity, that extensive tissue damage has taken place. As a third guide to the extent of the injury we have found complete displacement of the fragments also to be of importance. This relationship between the extent of the injury and the speed of bone union exists even when other factors, such as the method of treatment employed, are taken into consideration. In the present study it proved to be the most important single factor determining rate of fracture union. No investigation of other variables or comparison of results of different techniques of treatment can be considered valid unless the severity of the initial injury is taken into consideration.

The effects of traction and distraction on speed of union—Skeletal traction is frequently condemned because of its alleged retarding effect on the speed of fracture union. Albert (1944) and Watson-Jones and Coltart (1943), in studies of tibial fractures collected from a number of different centres, found that those treated by simple immobilisation united, on the average, more rapidly than those treated by skeletal traction, and they attributed this difference to

the effects of the traction. However, many surgeons reserve skeletal traction for cases of severe comminution or extensive compound wounding (Sherman 1940, Venable and Stuck 1947). Without allowing for possible variations in the severity of injury between the two groups, no firm conclusion can be derived from any differences that may be found in their average speed of union.

There seems to be no doubt, from our own study, that delay in healing takes place if traction causes distraction of the bone ends. If distraction is avoided, however, the union of fractures of moderate severity appears to be unaffected by traction. Here our results are at variance with the controlled study of closed tibial fractures reported by Landoff in 1948. In severe fractures we found the average time of union somewhat longer in cases subjected to traction, although there was no increase in the incidence of delayed union or non-union.

It may be concluded from this study that should skeletal traction be indicated in a particular fracture it may be used without risk of precipitating serious delay in healing, provided distraction is avoided.

SUMMARY

1. A series of 343 tibial shaft fractures proceeding to sound union in adults, and 192 fractures in children, was studied. Groups of fractures differing from each other only in the one particular variable under consideration were compared.

2. The severity of the injury (as assessed by the degree of displacement, of comminution and of compound wounding) was found to be an important determinant of speed of fracture union and of incidence of delayed and non-union. A simple classification of severity of injury is described.

3. Distraction delayed healing. Traction which avoided distraction had no effect on the rate of union of fractures of moderate severity compared with cases of similar severity of injury treated by immobilisation only. Major fractures subjected to traction did take longer to unite on the average, but there was no increase in the incidence of delayed or non-union.

I would like to thank Mr F. W. Holdsworth for permission to examine the patients in this study, all of whom were under his care, and also for his advice and interest during the preparation of this paper. Dr C. W. Wilson kindly confirmed the statistical significance of the figures used in constructing the tables.

REFERENCES

ALBERT, M. (1944): Delayed Union in Fractures of the Tibia and Fibula. Journal of Bone and Joint Surgery, **26,** 566.

CARPENTER, E. B., DOBBIE, J. J., and SIEWERS, C. F. (1952): Fractures of the Shaft of the Tibia and Fibula. Archives of Surgery, **64,** 443.

ELLIS, H. (1956): A Study of Some Factors Affecting Prognosis Following Tibial Shaft Fractures. Thesis, Bodleian Library, Oxford.

GRIFFITHS, D. Ll. (1942): Delayed Union of Fractures. Lancet, **ii,** 10.

LANDOFF, G. A. (1948): A Comparative Study of Methods of Treatment of Diaphyseal Fractures of the Leg. Acta Orthopaedica Scandinavica, **18,** 37.

SHERMAN, W. O. (1940): Treatment of Compound Fractures. Archives of Surgery, **40,** 838.

STARR, K. W. (1947): The Causation and Treatment of Delayed Union in Fractures. London: Butterworth & Co. (Publishers) Ltd.

VENABLE, C. S., and STUCK, W. G. (1947): The Internal Fixation of Fractures. Oxford: Blackwell Scientific Publications Ltd.

WATSON-JONES, R., and COLTART, W. D. (1943): Slow Union of Fractures. British Journal of Surgery, **30,** 260.

WILSON, H. (1942): Delayed Union of Fractures. Lancet, **ii,** 9.

NEUROBLASTOMA IN ORTHOPAEDIC PRACTICE

P. I. BUSFIELD, LONDON, ENGLAND

From the Department of Surgery, Postgraduate Medical School, London

This paper is intended to bring attention to the frequency with which cases of neuroblastoma may present as orthopaedic problems. The subject has attracted little attention in orthopaedic literature. Recent advances in treatment, with an improvement in the prognosis, justify a review of the orthopaedic aspects of the disease.

PATHOLOGY

Neuroblastoma is a malignant tumour arising from cells which have their origin in the neural crest. In the early embryo cells migrate from the region of the developing spinal cord to form the ganglia of the sympathetic nervous system and the medulla of the adrenal glands. Neuroblastoma most commonly arises in the adrenals, but it may also arise in sympathetic tissue behind the peritoneum, in the posterior mediastinum, or in the neck.

The histological appearances of the tumour vary with the degree of differentiation. The undifferentiated tumour consists of cells which are of uniform appearance, with scanty ill-defined cytoplasm and a single spherical nucleus. Mitotic figures may be numerous, and there are no nerve fibrils. Identification of the undifferentiated tumours may be difficult, and they were formerly interpreted as sarcomata. When partly differentiated the cells develop small fibrils and tend to become arranged in a characteristic rosette. Willis (1953) described the complete rosette as an arrangement of twenty or thirty cells with their nuclei at the periphery, surrounding a central area in which are contained the fibrils from each cell. It can be demonstrated that each fibril represents a small axon cylinder in continuity with the cytoplasm of the parent cell; the axon is non-myelinated and there are no neurilemmal cells. Complete differentiation is seen in the benign ganglioneuroma. This tumour contains adult nerve cells with mature nerve fibres which may be sheathed in neurilemma.

The gross appearance of the tumour is also variable. The adrenal neuroblastoma often appears to be confined within a capsule, showing no tendency to local infiltration. Less commonly there is widespread infiltration into the surrounding tissues. A remnant of the adrenal cortex may still be visible. Large areas of haemorrhage into the substance of the tumour are common, and calcification can often be demonstrated.

The tumour metastasises by the lymphatic route and by the blood stream. The paravertebral glands become extensively involved in abdominal neuroblastoma, and from there the tumour may infiltrate directly into the vertebral bodies. There may be extensive involvement of the cervical, axillary and inguinal lymph nodes. Haematogenous spread occurs most often in the liver, which may be grossly enlarged by discrete secondary deposits. Metastases have been recorded in all bones except those of the hands and feet, but they are most often found in the skull, pelvis, spine and the long bones. They occur occasionally in the skin. Gross involvement of the lungs is uncommon.

The standard text-books emphasise two clinical syndromes which have become the classical presentation of neuroblastoma. The Pepper syndrome comprises a primary adrenal tumour with metastases in the liver. The Hutchinson syndrome is produced by secondary deposits in the orbit and the skull. The literature of the last five years confirms that this presentation is incomplete. An analysis of the cases reported shows that a considerable proportion include features of particular concern to orthopaedic surgeons.

DISTRIBUTION AND INCIDENCE

The clinical series reported in the last five years have been reviewed. There were 177 cases of neuroblastoma in which the site of the primary tumour was stated. They were distributed as follows: 1) retroperitoneal primary 135 (76 per cent); 2) intrathoracic primary eighteen (10·2 per cent); 3) primary in the neck nine (5·1 per cent); 4) primary elsewhere fifteen (8·5 per cent). These last include a variety of sites—the pelvis, the naso-pharynx, the central nervous system and the internal ear.

There were ninety-six patients with neuroblastoma in which the sex of the patient was stated. They were equally divided—forty-eight were male and forty-eight female.

There were 104 patients with adrenal neuroblastoma in which the side of the primary tumour was stated. Sixty (57·7 per cent) were on the right side and forty-four (42·2 per cent) on the left. This is not statistically significant.

In no case was the primary tumour bilateral. In ninety-six cases the age of the patient was stated. Fifty-two (54·2 per cent) were under two years. Most of the others were children under ten years. The disease is rare in adults but several cases have been reported.

DIAGNOSIS

In any uncommon disease the presenting symptoms are of particular importance because they will usually determine to which hospital department the patient will be referred. This in turn will allocate the responsibility of diagnosis, a responsibility that is heaviest when early diagnosis can influence the outcome of the disease.

In the series reviewed there were 106 cases in which the presenting symptoms were recorded. They fall into four distinct groups.

Group 1. *Abdominal* (*fifty-one-cases*—47 *per cent*)—Attention was immediately directed to the abdomen. Abdominal pain, distension and vomiting were the commonest symptoms. In some patients a mass was felt on routine examination. A few presented with abdominal pain which resembled acute appendicitis.

Group 2. *Orbital* (*nine cases*—8·5 *per cent*)—In these patients the presenting symptoms were proptosis of one or both eyes, spontaneous haemorrhages into the eyelids, or deterioration in vision.

Group 3. *General* (*twenty-six cases*—24 *per cent*)—Here the presenting features were the general symptoms of malignant disease in childhood. These symptoms include lethargy, loss of weight and a diminishing appetite and energy. Unexplained pyrexia and a mild anaemia were commonly present.

Group 4. *Orthopaedic symptoms* (*twenty-four cases*—22·5 *per cent*)—In these patients the presenting symptoms were such that the advice of an orthopaedic surgeon might have been sought. They included pain about a joint, usually in the lower limbs; an unexplained limp or a disinclination to walk; acute pain in a bone, with signs resembling acute osteomyelitis; backache; and the development of paraplegia.

It appears from this analysis that almost a quarter of the patients with neuroblastoma will present as orthopaedic problems. It is characteristic of malignant disease in children that the early symptoms may regress spontaneously. Furthermore, when orthopaedic symptoms first appear they may not be accompanied by any radiological abnormality. The diagnosis is usually not suspected until either proptosis or a palpable abdominal tumour appears, and a number of weeks may pass before either of these is noticed. These circumstances combine to make early diagnosis difficult unless the condition is suspected and a full investigation carried out on suspicion alone.

Clinical examination—The detection of an abdominal swelling is the most important physical sign. The swelling may be due to the primary tumour, to the displaced kidney or to enlargement of the liver. It may be high under the costal margin and difficult to feel, or it may be a mass which almost fills the abdomen. Ascites is occasionally present, and there may be engorgement

of the veins of the abdominal wall. Vena caval obstruction may lead to oedema of the lower limbs. The eyes should be closely examined for proptosis, and the lids and conjunctivae for areas of haemorrhage. Palpation of the scalp may reveal subperiosteal nodules. A full neurological examination is important, with particular reference to power, sensation and reflexes in the lower limbs.

The axillary, cervical and inguinal lymph nodes should be palpated.

Special investigations. *Radiography of the abdomen*—There may be a soft-tissue shadow caused by the tumour or by the displaced kidney. Calcification commonly occurs in the tumour and may be demonstrable radiologically. It may appear as fine speckling in the suprarenal area or as a dense radio-opacity. The diaphragm may be elevated by the tumour or by the enlarged liver.

Pyelography—This is the most important single investigation. Usually it shows displacement of the renal pelvis downwards and laterally. Occasionally the pelvis is distorted by invasion of the kidney. Rarely there may be ureteric obstruction and hydronephrosis.

Radiography of chest—This may show a tumour in the posterior mediastinum or metastases in the lung fields. Secondary deposits may be seen in the ribs. In a " dumb-bell " tumour there may be destruction of vertebral transverse processes and adjacent parts of the ribs.

Radiography of long bones—Secondary deposits are usually osteolytic, and may be bilateral and symmetrically placed in the region of the metaphyses. Subperiosteal new bone may be laid down in laminated " onion-skin " layers. Occasionally there may be extensive localised destruction leading to pathological fracture.

Radiography of the skull—The calvarium may be stippled with areas of increased transradiance. Subperiosteal new bone may be laid down perpendicularly to produce " sun-ray " spiculation. Intracranial extension may cause increased intracranial pressure and widening of the sutures.

Examination of the blood—A mild anaemia is often present early in the disease. Severe anaemia suggests extensive involvement of the marrow. Leucocytosis is not uncommon. The white cell count was above 10,000 in three of the ten cases recorded in the Survey of Cancer in London (Harnett 1952).

Erythrocyte sedimentation rate—The only reference to an increase in the sedimentation rate is by Gross (1955). This is an important investigation which may give early warning of the sinister background to an apparently innocent symptom. It was elevated at some stage in each of the cases reported here and was an important early feature in Case 2.

Histological study of material obtained by sternal marrow puncture—This occasionally demonstrates tumour cells when the marrow is extensively involved. It has also been used to follow the effect of chemotherapy on metastases of microscopic size.

Histological study of biopsy specimens—This is not an infallible investigation. Considerable difficulty may be encountered in identifying an anaplastic tumour, and a haemorrhagic area may fail to yield malignant cells.

It has been our experience, and is the impression given by many of the cases reviewed, that when a patient presents with orthopaedic symptoms general examination usually does not reveal any reason to suspect the diagnosis. There are a few clinical syndromes which should immediately arouse suspicion. A case of seeming osteomyelitis with comparatively little elevation in the white cell count, with mild anaemia and a raised sedimentation rate, should suggest neuroblastoma. The development of paraplegia in a child, with no radiological changes in the spine, means that neuroblastoma must be excluded. Spontaneous haemorrhages along the margins of the eyelids warrant extensive investigation.

CASE REPORTS

Case 1—A boy aged eleven years presented in 1956 with a history of pain in the lumbar region which had started a few weeks before. About the same time it was noticed that bruising appeared spontaneously in the right upper eyelid. He began to walk with a limp and his parents

noticed that he seemed to tire easily. On first examination it was noted that there was some wasting of the right quadriceps; the right knee-jerk was absent, and there was an area of impaired sensation below the right knee. There were no other physical signs, and radiographs of the thoracic and lumbar spine appeared normal.

Two months later proptosis of the right eye was noticed and the lower pole of the right kidney became palpable. By that time extensive paresis of the right lower limb and anaesthesia over the third, fourth and fifth lumbar dermatomes had appeared. The intravenous pyelograph showed a deformity of the right renal pelvis. Sternal marrow smear was suggestive of neuroblastoma.

FIG. 1

Case 1—The primary tumour. The tumour has been cut across and the kidneys bisected. The tumour appears to be encapsulated and there are numerous areas of haemorrhage. Some of the involved para-aortic glands are shown in the specimen.

After a further month the patient had a complete flaccid paraplegia with anaesthesia below the fifth thoracic level and a paralysed bladder. The sedimentation rate was then 70 millimetres in the first hour. The haemoglobin was 52 per cent and the white cell count was 4,200 per cubic millimetre. Radiographs of the skeleton showed widespread deposits in both humeri and femora, and in the pelvis, but no evidence of vertebral metastases.

Irradiation was given to the region of the right orbit. There was a severe reaction which resulted in opacity of the cornea, but the proptosis subsided. A course of oral and intravenous radioactive phosphorus was given but no response was apparent. His general condition deteriorated despite

repeated blood transfusions and he died seven months after the onset of symptoms. The diagnosis was confirmed at necropsy. The primary tumour is shown in Figure 1.

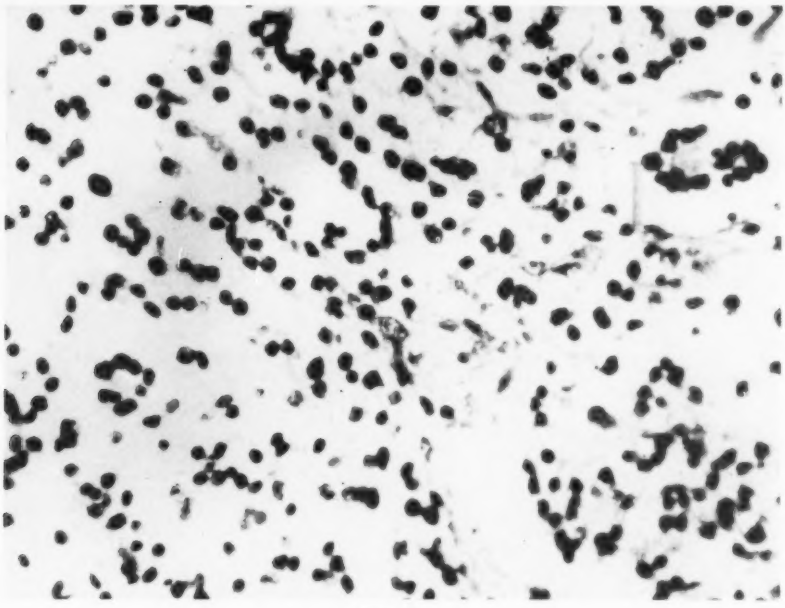

FIG. 2
Case 2—Section of the primary tumour showing the incomplete formation of rosettes.

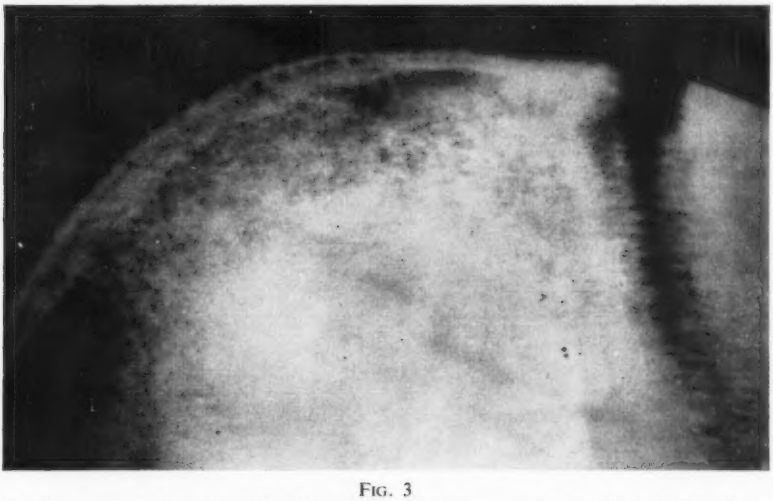

FIG. 3
Case 2—Part of the radiograph of the skull. This demonstrates the " stippling " of the calvarium, the formation of " sun-ray " spicules and widening of the sutures, which together are diagnostic of neuroblastoma.

Case 2—A boy aged five years presented in 1955 with a history of pain in the left hip which was attributed to a blow sustained a few days before. Examination at that time showed tenderness over the front of the hip joint and slight limitation of rotation. There were no other physical signs.

The Mantoux reaction was negative. The haemoglobin was 70 per cent and the white cell count was 10,000 per cubic millimetre. The sedimentation rate was 45 millimetres in the first hour. Radiographs of the hip were normal.

The hip was treated by continuous traction and the physical signs disappeared. There was, however, evening pyrexia, on occasions to 101 degrees Fahrenheit, and the sedimentation rate remained high. His general condition slowly deteriorated, and he began to complain of pain and stiffness in the neck. The left knee became painful and swollen, but there was no abnormality on the radiographs. He developed fleeting pains in other joints, and rheumatic fever was suspected. His symptoms appeared to subside with a course of salicylates, but the sedimentation rate remained over 40 millimetres.

Five months later a swelling was noted in the scalp, and biopsy of this enabled the diagnosis of neuroblastoma to be established. He rapidly developed numerous swellings in the scalp and generalised enlargement of cervical, inguinal and axillary lymph nodes. Irradiation was attempted, but was

FIG. 4

FIG. 5

Case 3. Figure 4—Radiograph of the right lower ribs. There is marked subperiosteal new bone formation.
Figure 5—Photograph showing the temporal swelling and proptosis.

discontinued because the general condition rapidly deteriorated. A course of Vitamin B 12 was started but the boy died three weeks later, seven and a half months after the onset of symptoms. A small mass became palpable in the left hypochondrium two weeks before his death. The diagnosis was confirmed at necropsy (Figs. 2 and 3).

Case 3—A boy aged twelve years presented in 1956 with a painful swelling over the right lower ribs which he attributed to an injury sustained a few weeks before. Radiographs showed extensive destruction of the tenth rib with subperiosteal new bone, and also a small pleural effusion (Fig. 4). The condition was regarded as osteomyelitis and treated with a course of penicillin. There was rapid improvement, but within a few weeks the swelling returned larger than before. Biopsy was performed three months after the onset of symptoms. The histological appearances appeared to confirm the diagnosis of osteomyelitis, and treatment was continued with acromycin. The sedimentation rate rose to 127 millimetres in the first hour, and later a swelling was noted on the lateral aspect of the left orbit. A further biopsy enabled the diagnosis of neuroblastoma to be made.

The patient subsequently developed generalised bone metastases and enlargement of the liver. Palliative irradiation was given. He died eight months after the onset of symptoms (Figs. 5 and 6).

Case 4—Boy aged five years. This boy presented in 1951 with a history of pain in the right arm of four days' duration. Examination at that time showed tenderness and thickening over the middle of the shaft of the humerus, with increase in local heat. General examination showed no other abnormality. The temperature was normal. The haemoglobin was 80 per cent and the white cell count was 8,500 per cubic millimetre. The sedimentation rate was 20 millimetres in the first hour. The radiographs showed a little patchy rarefaction with no periosteal reaction.

Open biopsy was carried out. The bone was found to contain material which resembled thick, yellow pus. The histological appearance suggested secondary neuroblastoma. Soon a swelling became palpable in the abdomen, and the intravenous pyelograph demonstrated downward displacement of the right kidney.

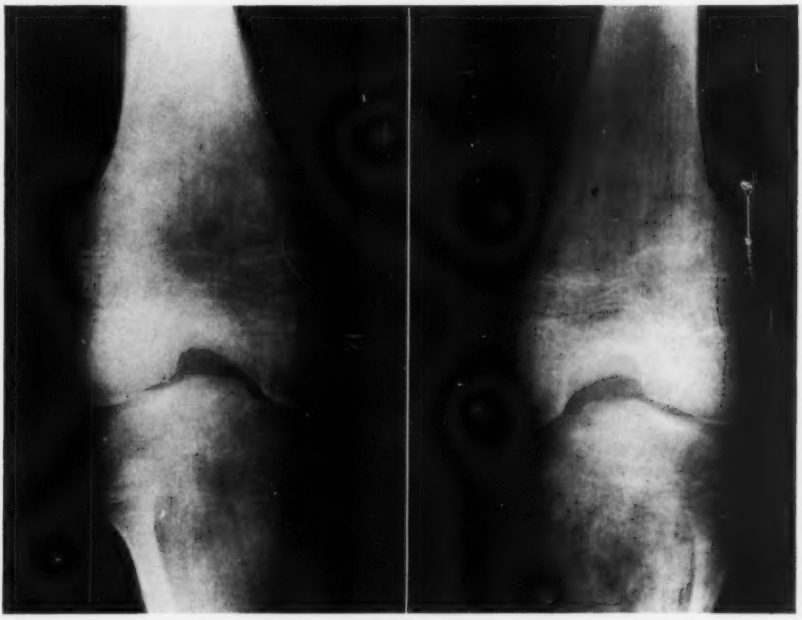

FIG. 6

Case 3. Radiographs showing the characteristic arrangement of bone metastases in neuroblastoma. The metastases may be bilateral, symmetrical and situated in the region of the metaphyses. They usually appear late in the disease.

The lesion in the humerus extended and pathological fracture occurred. Palliative irradiation was given. The child died nine months after the onset of symptoms.

Case 5—A girl aged sixteen years presented in 1955 with a history of pain over the anterior part of the left iliac crest; this had been increasing in severity over the previous three months. She had sustained a minor injury to the inguinal region on the same side a few weeks before the onset of the pain. Before admission to hospital she had been treated with penicillin without improvement.

Examination at that time showed tenderness over the anterior inferior iliac spine. The radiographs demonstrated a little subperiosteal new bone in the area of tenderness. The patient was treated with continuous traction and a course of streptomycin, and her symptoms improved. The sedimentation rate was persistently raised to between 30 and 40 millimetres in the first hour. The white cell count varied between 6,000 and 12,000 per cubic millimetre. Her temperature remained elevated, on occasions to 100 degrees Fahrenheit.

A month later she was transferred for further investigation. There was now a tender swelling on the outer aspect of the left ilium. The radiographs showed increased new bone formation, with underlying areas of increased density and rarefaction. The sedimentation rate was now 90 millimetres

in the first hour, and the haemoglobin was 88 per cent. There were no other physical signs. Radiographs of other bones showed no abnormality.

Open biopsy was performed and histological examination of the specimen established the diagnosis of neuroblastoma. The intravenous pyelograph appeared to be normal. Palliative irradiation was given to the ilium, with considerable symptomatic relief, and she was allowed to return home. She died eight months after the onset of symptoms. The necropsy showed widespread metastases, and the primary tumour appeared to have arisen in the posterior mediastinum.

Case 6—Boy aged twelve years. This boy presented in 1951 with a history of pain and swelling in the right thigh which had started four months before. The pain had been intermittent but the swelling had slowly increased in size. Examination at that time showed a diffuse, hard swelling attached to the lowest third of the femur. There were no other physical signs. The radiographs showed a tumour in the soft tissues, and underlying this an area of subperiosteal new bone which was laminated. The appearance suggested either an osteogenic sarcoma or a Ewing's tumour. The haemoglobin was 71 per cent and the white cell count was 9,300 per cubic millimetre. The sedimentation rate was 25 millimetres in the first hour.

Open biopsy was performed and the histological appearance suggested either a Ewing's tumour or a secondary neuroblastoma. Radiographs of the rest of the skeleton showed no evidence of other metastases. The intravenous pyelograph showed that the left kidney was slightly displaced downwards. There was slight hydronephrosis but no distortion of the calyces. The twelfth rib on the same side was completely destroyed.

Because of the size of the secondary tumour in the femur amputation was performed through the upper third of the thigh. The patient made a good recovery and was symptom-free until a year later, when further lesions appeared in the mandible, the scalp and around the left knee. Radiographs of the abdomen now showed calcification in the left suprarenal area, destruction of the adjacent transverse processes and an area of calcification extending along the left side of the lower thoracic vertebral bodies. He died one year and nine months after the onset of symptoms.

DIFFERENTIAL DIAGNOSIS

The diagnostic problem presented to the orthopaedic surgeon is usually that of a local condition in bone combined with non-specific general manifestations. The conditions which have to be excluded fall into two groups: 1) local bone disease with general manifestations; and 2) general disease with localised manifestations in bone. Although the list of theoretical possibilities would be a long one, the conditions which, in practice, have to be differentiated are few.

Local bone disease. *Osteomyelitis*—The commonest difficulty arises when inadequate treatment with antibiotics has been given, with some apparent improvement. The association of a lowered haemoglobin with a raised sedimentation rate should lead to intensive investigation, including open biopsy, rather than a clinical trial with a different antibiotic.

Ewing's tumour—To avoid entering discussion on the pathogenesis of this tumour let it suffice to say that it may be difficult to differentiate between a primary tumour in bone and an apparently solitary metastasis from a neuroblastoma. It is important in such a case to obtain radiographs of the rest of the skeleton. Pyelography will often establish the diagnosis in adrenal neuroblastoma.

Osteogenic sarcoma—Although there may be some constitutional disturbance in bone sarcoma this is not usually as marked as in neuroblastoma. Skeletal radiographs, pyelography and biopsy will usually distinguish these tumours.

" *Occult*" *tuberculosis*—The early stages of bone tuberculosis, before radiographic changes are marked, may be difficult to distinguish from neuroblastoma. If the diagnosis is still in doubt after full investigation biopsy should be preferred to a clinical trial with streptomycin.

Generalised diseases. *Rheumatic fever*—Neuroblastoma may closely simulate the fleeting joint pains of rheumatic fever without radiographic changes in the early stages. A persistently raised sedimentation rate, despite symptomatic improvement, may be the only early sign.

Aleukaemic leukaemia—The distinction may be difficult in cases of neuroblastoma with early involvement of superficial lymph nodes. Sternal marrow puncture may afford the only conclusive proof.

TREATMENT AND PROGNOSIS

The treatment of neuroblastoma may be considered under three headings.

Surgery—Most authors in the series reviewed favoured a surgical attack on the primary tumour whenever practicable. The surgical ideal of excision of the primary growth with its regional lymphatic field is seldom possible. Good results are claimed in the cases in which it has been possible. Koop, Kiesewetter and Horn (1955) found six such cases in their series of thirty-nine and claimed that all have been cured. The proportion of their cases without distant metastases is unusually high.

There is wide difference of opinion on the procedure to be adopted when laparotomy shows that complete excision is impossible. In most cases only a biopsy specimen has been taken. Koop, Kiesewetter and Horn (1955) performed what they described as a " major surgical insult " to the tumour, removing as much as they could without regard to the accepted principles of cancer surgery. They claimed twelve cures out of thirty-nine cases.

Poore, Dockerty, Kennedy and Walters (1951) recorded a case in which there was lymphatic extension beyond the limits of excision, and in which they had to divide the inferior vena cava to remove the primary tumour. This patient has now survived for several years.

Irradiation—Deep x-ray therapy has been used to effect cure, as a post-operative prophylactic, and as a palliative. The tumour is usually radio-sensitive, and often remarkably so. Phillips (1953) delivers 2,000–3,000 r. over ten to twenty days to destroy an adrenal tumour. Uhlmann and von Essen (1955) record six three-year survivals out of twenty cases treated (30 per cent).

Irradiation is not without danger. Radiation nephritis is a recorded complication when high dosage is given, and one fatal case has been reported. Depression of marrow function has led to cessation of treatment in several cases, and has caused one death.

Opinion appears to be divided about the value of post-operative irradiation. Wittenborg (1950) reviewed seventy-three cases in which the treatment was by operation, irradiation or both. Twenty-two of the patients survived for three years or more. In all the survivors the disease was confined to the abdomen.

Chemotherapy—The results already quoted are unfortunately of little more than theoretical interest to orthopaedic surgeons because the patients they see will usually present with bone metastases. In almost all the cases reviewed those with bone involvement have died, and survivals have been rare in the past. Hansen (1953) reported a patient who had bilateral proptosis and a deposit in the mandible, who was treated with irradiation alone, and who was well after twelve years. Goldring (1951) reported a case in which bilateral proptosis was combined with multiple metastases in liver and bone. The primary tumour alone was irradiated. The child was alive fifteen months later; the bone secondaries had disappeared and the proptosis subsided.

The only form of treatment that offers hope of success in cases with bone involvement is some kind of chemotherapy. A number of substances have now been used. Nitrogen-mustard has been tried extensively without much success. Some authorities claim that it has a particular effect on secondaries of microscopic size. It may be given intravenously (0·4 milligram per kilogram of body weight). Folic acid antagonists have been used in recent years with some success. Phillips (1953) recommended amethopterin (2·5 milligrams daily for ten days) in repeated courses with intervals of fourteen days.

Interest has recently been centred on the use of vitamin B 12, which has been used at the Hospital for Sick Children, Great Ormond Street, for the last four years. The preliminary reports show that a quarter of the patients survived for more than two years without recurrence. In other cases there has been some improvement and a fatal issue has been delayed. The patients selected for treatment usually had widespread disease and could not otherwise have been expected to survive. One milligram of vitamin B 12 is given on alternate days and the dosage is maintained for two years or more. Dr Martin Bodian is preparing a detailed account of his experience with this form of treatment.

DISCUSSION

It seems that the principles of treatment in neuroblastoma are becoming established. The usual aim in all the recent series has been to attack the primary tumour even in the presence of metastases. It is widely felt that the primary tumour has some influence on the rate and extent of secondary spread, and that the metastases are more responsive to treatment when the primary has been controlled in some way.

A good deal of attention has been directed to the spontaneous " maturation " of the malignant neuroblastoma into the benign ganglioneuroma. A small number of cases have now been reported of prolonged survival when no active treatment has been given. The case of Cushing and Wolbach (1927) has been repeatedly quoted as a proven example of this process of maturation. In this case a biopsy of the extraspinal extension of a " dumb-bell " tumour demonstrated neuroblastoma. Ten years later the intraspinal extension was removed by Cushing and proved to be a benign ganglioneuroma. This has been interpreted as meaning that the intraspinal extension had undergone maturation. This interpretation is not necessarily correct, because it is well known that malignant and benign areas can co-exist in the same tumour. It seems more likely that the malignant tumour undergoes self-destruction by haemorrhagic necrosis, which in this case was induced by the biopsy. Whether or not one may accept the possibility of spontaneous cure is of more than theoretical importance. The tendency in recent years has been to treat these patients by radical methods even in the presence of metastases, and radical treatment might be withheld if spontaneous cure is considered to be even a remote possibility.

In the series reviewed there were seven cases of paraplegia. This may be spastic or flaccid, and the type of paralysis may be of some prognostic significance. Spastic paraplegia results from cord compression which may be due to a primary " dumb-bell " tumour, as in the case quoted above. A similar case is reported by Elefant, Jeklerova and Lesny (1955) in a baby of seven months, in whom the paraplegia was associated with a large paravertebral tumour, and the whole tumour was malignant. The tumour was treated by partial excision and post-operative irradiation. The child was reported to have survived for two years without recurrence and with considerable recovery of function. The cause of flaccid paraplegia is suggested by the post-mortem findings in Case 1. There was extensive involvement of the paravertebral glands with contiguous extensions within the spinal canal. The spinal cord was destroyed from the level of the fifth thoracic vertebra downwards. It seems that the cord lesion had resulted from interference with the blood supply of the cord, as there was no sign of cord compression.

SUMMARY

1. Neuroblastoma is a disease that may concern the orthopaedic surgeon closely.

2. Six such cases are reported.

3. Early diagnosis may be difficult, but can be achieved if the condition is suspected and an adequate investigation is carried out on suspicion alone.

4. The prognosis in these patients is still poor, but might be improved by a vigorous combination of surgery, irradiation and chemotherapy. The recent advance in treatment with vitamin B 12 may improve the prognosis substantially.

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REFERENCES

BODIAN, M., and WHITE, L. L. R. (1953): Preliminary Observations on the Treatment of Neuroblastoma with Vitamin B 12. Annual Report of British Empire Cancer Campaign, **31,** 174.

BODIAN, M., and WHITE, L. L. R. (1954): Neuroblastoma. A General Review. Report of British Empire Cancer Campaign, **32,** 195.

BODIAN, M., and WHITE, L. L. R. (1955): Neuroblastoma and Vitamin B_{12} Therapy. Report of British Empire Cancer Campaign, **33,** 186.

BODIAN, M. (1956): Eighth International Congress of Paediatrics, Copenhagen.

BODIAN, M. (1957): Personal communication.

CHARACHE, H. (1954): Neuroblastoma. A Report of Ten Cases and Review of the Literature. American Journal of Surgery, **87,** 545.

CUSHING, H., and WOLBACH, S. B. (1927): The Transformation of a Malignant Paravertebral Sympathicoblastoma into a Benign Ganglioneuroma. American Journal of Pathology, **3,** 203.

ELEFANT, E., JEKLEROVA, J., and LESNY, I. (1955): Rückenmarkskompression im Säuglingsalter. Annales Paediatrici, **184,** 253.

GOLDRING, D. (1951): Neuroblastoma Sympatheticum with Metastases. Report of a Case with Apparent Recovery. Journal of Pediatrics, **38,** 231.

GROSS, H. (1955): Ein kasuistischer Beitrag zum Neuroblastoma sympathicum. Wiener klinische Wochenschrift, **67,** 681.

HANSEN, P. B. (1953): Sympathicoblastoma of the Adrenal Medulla with Osseous Metastases. Acta Radiologica, **40,** 500.

HARNETT, W. L. (1952): Survey of Cancer in London. London: British Empire Cancer Campaign.

IMMERMANN, E. W., and COMPERE, E. L. (1954): Bone Metastases of Neurogenic Tumors of the Sympathetic Nervous System in Children. Quarterly Bulletin of the Northwestern University Medical School, **28,** 138.

KOOP, C. E., KIESEWETTER, W. B., and HORN, R. C. (1955): Neuroblastoma in Childhood. Survival After Major Surgical Insult to the Tumor. Surgery, **38,** 272.

OBERKIRCHER, O. J., STAUBITZ, W. J., and PARMENTER, F. J. (1953): A Clinical Study of Neuroblastoma. Journal of Pediatrics, **43,** 177.

PHILLIPS, R. (1953): Neuroblastoma. Hunterian Lecture. Annals of the Royal College of Surgeons of England, **12,** 29.

POORE, T. N., DOCKERTY, M. B., KENNEDY, R. L. J., and WALTERS, W. (1951): Symposium on Surgical Aspects of the Cancer Problem-Abdominal Neuroblastomas. Surgical Clinics of North America, **31,** 1,121.

REIQUAM, C. W., BEATTY, E. C., Jun., and ALLEN, R. P. (1956): Neuroblastomas in Infancy and Childhood. A.M.A. Journal of Diseases of Children, **91,** 588.

UHLMANN, E. M., and ESSEN, C. von (1955): Neuroblastoma (Neuroblastoma Sympatheticum). Pediatrics, **15,** 402.

WILLIS, R. A. (1953): Pathology of Tumours. Second edition. London: Butterworth & Co. (Publishers) Ltd.

WITTENBORG, M. H. (1950): Roentgen Therapy in Neuroblastoma. Radiology, **54,** 679.

HYPERKALAEMIA IN TRAUMATIC RENAL FAILURE

Report of Two Cases

KENNETH S. MORTON and FRANK P. PATTERSON, VANCOUVER, CANADA

From the Department of Orthopaedic Surgery, Vancouver General Hospital, and the Faculty of Medicine, University of British Columbia

Surprisingly little mention has been made in the orthopaedic literature of the elevation of serum potassium in post-traumatic acute renal failure. The importance of this complication of trauma is indicated by Teschan, Post *et al.* (1955) in quoting Mallory's figure of 18·6 per cent renal deaths in 427 battle casualties; Simeone's of 14 per cent of deaths in 165 necropsies in Korean casualties; and a group of patients severely wounded in the second world war of whom 40 per cent developed acute renal failure. These authors themselves found, in 8,000 Korean casualties, an incidence of acute renal failure of 0·5 per cent; but in a severely wounded group of forty-three casualties this incidence rose to 21 per cent. The frequency and rapidity of development of potassium intoxication was striking in their cases; death from cardiac arrest of potassium intoxication occurred as early as the fourth day. Of seventy-two dialyses carried out in treatment, fifty were done for hyperkalaemia alone or for hyperkalaemia with uraemia (Smith, Post *et al.* 1955).

That the condition is also encountered in civilian practice is recorded by Swann and Merrill (1953), who summarised current knowledge of acute renal failure and stated their experience with eighty-five cases. Hyperkalaemia occurred in most of their cases, and frank potassium intoxication in 25 per cent.

Recently we have encountered two cases in which potassium rose to alarmingly high levels, such that death from its cardiotoxic effect was threatened. In both cases the question of surgical treatment by evacuation of a large haematoma or ligation of large vessels from a damaged limb, as ready sources of free potassium, was debated.

CASE REPORTS

Case 1—A man aged thirty-two years suffered right above-knee and left below-knee traumatic amputations in a logging accident. First-aid and resuscitative measures were used before his admission to the Vancouver General Hospital on the third day. Oliguria was already apparent, the urine output averaging 500 cubic centimetres a day for these three days. He was conscious but confused. There was moderate oedema of the perineum, and marked swelling and ecchymosis of the right flank, hip and thigh. The blood pressure was 110/90 millimetres Hg., pulse 96 per minute, haemoglobin 14·8 grammes per cent. The potassium was already elevated to 6·5 m.Eq. per litre. Gas gangrene and tetanus antisera, penicillin and streptomycin were given. The mental state fluctuated between clarity and confusion. Urine output continued to be low, averaging 725 cubic centimetres for the next three days with an oral and parenteral fluid intake approaching two litres daily. Significantly, the specific gravity of the urine was recorded as 1·008 and 1·006 on two occasions and contained gross blood and 3 plus protein. Electrolyte determinations showed hypochloraemia and hyperkalaemia, and uraemia was evident (Table I). On the fifth day after the injury the blood pressure rose to 140/90 millimetres Hg. and the following day an attempted aspiration of the haematoma of the right flank failed. The original stump dressings were removed under cyclopropane anaesthesia. Both stumps required further cleansing and debridement and were packed open. Thrombosed anterior and posterior tibial vessels were noted in the below-knee stump. One litre of matched blood was given during the procedure, and the blood pressure remained between 130/75 and 160/90 millimetres Hg.

Immediately after the operation the patient's general condition improved and he was mentally clear, with a blood pressure of 150/85 millimetres Hg. and a pulse of 110 per minute. The next day he was restless, developed a cough, and was irrational. He became more difficult to nurse and, when turned on his side, became suddenly cyanotic and died, twenty hours after operation.

At necropsy, performed the day after death, there was a large haematoma in the right abdominal muscles, extending into the right thigh and across the pubis. All the superficial and deep veins were thrombosed except for their upper parts, which were free of clot.

TABLE I

BIOCHEMICAL INVESTIGATIONS IN CASE 1

Day after injury	Urine output (*cubic centimetres*)	Na m.Eq. per litre	Cl m.Eq. per litre	CO_2 m.Eq. per litre	K m.Eq. per litre	NPN (*milligrams per cent*)	Creatinine (*milligrams per cent*)
3	?405	129	81	34	6·5	76	4·4
4	690	132	82	29	6·1	111	5·8
5	750	130	78	32	6·1	123	6·7
6	725	134	84	25	6·2	123	7·2
7	400						

The femoral arteries were thrombosed and there were irregular ischaemic changes in the muscles of the left leg. *The lungs* showed a recent organised thrombus partly obstructing the pulmonary artery and extending back into the right ventricle and auricle. *The kidneys* weighed 600 grammes and cortical pallor was noted. *Microscopically*, the kidneys were described as typical of the so-called crush or shock kidney, with disruption of the tubules, eosinophilic granular debris in the capsular space and tubules, and tubular rupture with interstitial reaction. The pathological diagnoses were massive pulmonary embolus due to phlebothrombosis and thrombophlebitis of the right leg and renal tubular degeneration and disruption with early cortical necrosis.

Comment—Though this patient was not severely oliguric, the non-protein nitrogen and creatinine were elevated to 123 milligrams per cent and 7·2 milligrams per cent respectively, and he was undoubtedly in acute renal failure in consequence of the severe injury and subsequent prolonged hypotension. Urine volumes were, in fact, frequently in the neighbourhood of 600 to 700 cubic centimetres, but this urine was of poor quality, the specific gravity being only 1·006 to 1·008. At such specific gravities, such volumes are entirely inadequate in disposing of toxic materials. A large haematoma in the right hip region was apparent, and when the serum potassium became elevated to 6·5 m.Eq., the surgical evacuation of the potassium-laden mass was considered. Risk of infection delayed this procedure, and when the kalaemia levelled off at 6·1 m.Eq., operation was felt to be unnecessary, though aspiration was attempted.

Case 2—A man aged twenty-two years was injured in a logging accident in 1954. When he was admitted to the Vancouver General Hospital three and a half hours after the injury his blood pressure was 60/35 millimetres Hg., and the pulse 90 per minute. His left foot and leg lay in lateral rotation and the left thigh was much enlarged and tense. Sensation was absent at the left foot; a faint anterior tibial artery pulsation was described but the foot was pale and cyanotic.

Haemoglobin was 7·7 grammes per cent, haematocrit 28. Intravenous Subtosan and whole blood raised the blood pressure to 85/45 millimetres Hg. within half an hour, and to

105/65 millimetres Hg. four hours after admission, but the pulse remained elevated to 120 per minute. Exploration of the left thigh vessels was considered, but, though the thigh became further enlarged and pulsation was not palpable, the foot was warmer and venous filling was noted; the operation was therefore not carried out. Routine antibiotics and antisera were given. No urine was obtained by catheterisation. On the first day after the injury he began vomiting, and the local swelling increased. Sensation was absent below the knee, and the left foot was cool. The urine output was 660 cubic centimetres. The specific gravity was not recorded, but examination for myoglobin was negative and for protein positive. The blood pressure had become constant at 120/70 millimetres Hg.

On three occasions 15 cubic centimetres of 2 per cent Xylocaine were injected through an epidural catheter to improve the blood supply to the limb. The blood pressure fell as low as 80/60 millimetres Hg. after the first of these injections. Ischaemic changes progressed, the foot became cyanotic and mottled, and the calf was swollen and hard. Non-protein nitrogen was elevated to 85 milligrams per cent and potassium to 7·2 m.Eq. (Table II). On this and the following days the fluid intake approached three litres daily.

TABLE II

BIOCHEMICAL INVESTIGATIONS IN CASE 2

Day after injury	Urine output (*cubic centimetres*)	Na m.Eq. per litre	Cl m.Eq. per litre	CO_2 m.Eq. per litre	K m.eq. per litre	NPN (*milligrams per cent*)
0						
1	660	140	96	28	7·2	85
2	330			30		83
3	580	132	90	33	9·2	111

On the second day the patient's condition remained static. The urine volume fell to 330 cubic centimetres with a specific gravity of 1·017. There was developing gangrene of the left foot. On the third day he became restless, hyperexcitable and confused; a sudden episode of dyspnoea and collapse responded well to intravenous Coramine and oxygen. An electrocardiograph showed changes characteristic of gross potassium intoxication. Potassium at this time was 9·2 m.Eq. and non-protein nitrogen 111 milligrams per cent. Consideration was given to ligation of the large vessels of the left thigh in an attempt to reduce the source of noxious substances. As an alternative, extra-corporeal dialysis was begun on the evening of the third day and continued for three hours.

Electrocardiographs during this period showed a progressive reversal of the potassium effect after seventy-five minutes of dialysis. The patient's general condition improved at first and the urine output increased, but ten hours after dialysis he suddenly collapsed and died on the morning of the fourth day after the injury.

At necropsy, performed the day of death, the right mid-thigh circumference was eighteen inches, the left twenty-seven inches. There was a fracture of the left femoral shaft, and the medial and posterior thigh muscles were completely stripped from the length of the femur. A cavity fifteen inches long and four inches in diameter, filled with blood, was thus formed. The femoral artery was torn at the supracondylar level and was constricted above and below this point. The femoral vein was ruptured just proximal to the tear in the artery.

Examination of the *heart* showed both ventricles to be dilated. The *kidneys* weighed 480 grammes, the cut surface bulged and the cortex was pale. *Microscopically*, cardiac muscle fibres were swollen, with some fragmentation. The kidney: congested glomerular tufts with increased capsular space; eosinophilic granular debris in this space and the tubules; loss of

lining epithelium and rupture of tubules; interstitial reaction with mononuclear and foreign body giant cells.

The final pathological diagnosis was multiple pulmonary emboli from phlebothrombosis and thrombophlebitis in veins of the left leg, and " shock kidney."

Comment—This patient became oliguric and uraemic after the injury, which lowered the blood pressure to below 100 millimetres Hg. (systolic) for a period of seven hours. A large source of potassium was evident clinically and at necropsy in the severely damaged thigh, and that potassium ions were in fact being freed into the circulation was evident from the characteristic electrocardiographic changes and biochemical determinations.

Less evident in this case is the fact that some degree of acute renal failure adds to the elevation of serum potassium. Though the urine volume fell as low as 330 cubic centimetres and the non-protein nitrogen rose to 111 milligrams per cent, the specific gravity of the urine remained at 1·011 to 1·017. Nevertheless it is apparent that the two factors, increased source of potassium and decreased excretion of it by a failing kidney, contribute to the hyperkalaemia. Swann and Merrill (1953) stated that acute renal failure connot be ruled out by a urine specific gravity of 1·015 to 1·020 during the first week, and in some of their cases daily urine volumes were 400 to 800 cubic centimetres. There seems little doubt, therefore, that these kidneys were failing, and it seems possible that early radical sacrifice of the limb might have lessened the eventual hyperkalaemia. Such surgery, however, would be a difficult action to take when it is considered that, on the basis of ischaemic changes, a below-knee amputation would have sufficed. It is unlikely that ligation of vessels or late radical amputation would have affected the course of the illness favourably. Notable was the dilated heart found at necropsy; this is characteristic of the cardiotoxic effect of potassium but also of an overloaded heart, here possibly the result of dialysis.

DISCUSSION

That potassium has a cardiotoxic effect has been shown by Winkler, Hoff and Smith (1941), and the electrocardiographic changes in hyperkalaemia have been described by Hoff, Smith and Winkler (1941), Derot, Pignard, Touraine and Bernard (1953), and Strauss (1948). Hoff *et al.* also pointed out that serum potassium does not reach this toxic level in experimental acute renal failure nor in chronic renal failure because of the high clearance of potassium. In fact, Muirhead and Hill (1948) stated that a daily intake of 0·5 to 1·0 gramme of potassium during the renal insufficiency phase of acute renal failure will not produce hyperkalaemia, and Platt (1953) found that, even when the glomerular filtration rate was reduced to 20 per cent of normal, the daily excretion of potassium remained normal.

Smith (1951) also referred to the fact that potassium is completely filtrable. Nevertheless, the cardiotoxic effect of potassium has been consistently described by several authors (Allen 1952; Balch, Meroney and Sako 1955; Burnett *et al.* 1947; Derot *et al.* 1953; Hicks, Crutchfield and Wood 1950; Humphrey and Avery Jones 1947; Knowles and Kaplan 1953; Stock 1949; Strauss 1948) as a precipitating cause of death in many cases of uraemia.

It seems necessary, therefore, to look for an intrinsic source of excess potassium to account for these clinical observations: a ready source is the intracellular component of body fluid. The intracellular concentration of potassium has been determined to be 115 to 160 m.Eq. per litre (Derot *et al.* 1953) and much can be released into the circulation by injury (Green 1945), prolonged ischaemia (Rewell 1943, Meroney and Herndon 1954) and catabolic processes (Balch, Meroney and Sako 1955; Derot *et al.* 1953). The small daily quantity of potassium from catabolism is easily excreted and may accumulate for weeks in anuria without reaching toxic levels. But in post-traumatic conditions there is excessive loss from damaged cells, devitalised tissue giving up potassium to the plasma (Fenn 1940).

Detection of dangerously elevated potassium levels must rely entirely on biochemical determinations and electrocardiographic examinations. The electrocardiograph shows the effects of potassium intoxication first (Meroney and Herndon 1954, Teschan *et al.* 1955,

Keith and Burchell 1949), and clinical signs such as paraesthesiae and flaccid paralysis appear only as terminal manifestations. It should be noted, too, that the cardiotoxic effects of potassium become apparent at variable serum levels, being dependent also on the calcium, phosphate and sodium levels (Meroney and Herndon 1954, Teschan *et al.* 1955).

One facet of the successful management of acute renal failure would, therefore, appear to be the control of hyperkalaemia. Early attention, in this regard, was paid to removal of the source by primary amputation, fascial incisions and gradual tourniquet release from pressure (Belsey and McMichael 1942), but recently medical means have been employed to remove excess potassium already in circulation. These have included extracorporeal and corporeal dialysis (Balch *et al.* 1955; Hicks *et al.* 1950; Meroney and Herndon 1954; Smith *et al.* 1955), cation exchange resins (Derot *et al.* 1953; Knowles and Kaplan 1953; Meroney and Herndon 1954) and hypertonic glucose with insulin (Allen 1952; Derot *et al.* 1953; Meroney and Herndon 1954; Smith *et al.* 1955). In one of the two cases reported above, dialysis in the Kolff dialyser was successful in relieving the potassium effect on the heart, as shown by reversal of the electrocardiographic changes. However, the patient died in spite of this success, possibly from multiple pulmonary emboli or pulmonary oedema. Dialysis is notably not without danger.

In both cases the question of surgical removal of a large source of potassium was raised. Smith *et al.* (1955), in their review of fifty-one cases of renal insufficiency in Korean battle casualties, concluded that radical debridement, early amputation of gangrenous parts and drainage of infected foci were essential, often blunting the rapid progress of uraemia. The danger of infection and of further shock is not negligible in the evacuation of a large haematoma and it seems unlikely that ligation of the larger vessels to and from an irreparably damaged limb would do any more than induce more ischaemia, with increased return of potassium to the main circulation through the smaller anastomotic vessels and by diffusion. Radical amputation would therefore have been the only logical procedure in Case 2, and such an operation is a formidable one in an already dangerously ill patient.

Nevertheless, decreasing the source of excess potassium by surgical or medical means is suggested as an essential part of the management of cases of traumatic renal failure with hyperkalaemia.

SUMMARY

1. Two cases are reported in which hyperkalaemia was a prominent feature in acute renal failure and in which obvious sources of excess potassium were apparent.
2. The literature pertaining to potassium in acute renal failure is briefly reviewed.
3. A return to the surgical elimination of large and expendable tissue sources of potassium in the management of acute renal failure with hyperkalaemia is suggested.

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REFERENCES

ALLEN, A. C. (1952): The Kidney, p. 72. London: J. and A. Churchill Ltd.
BALCH, H. H., MERONEY, W. H., and SAKO, Y. (1955): Observations on the Surgical Care of Patients with Post-traumatic Renal Insufficiency. Surgery, Gynecology and Obstetrics, **100,** 439.
BELSEY, R., and MCMICHAEL, J. (1942): *In* Discussion on the Effects on the Kidney of Trauma. Proceedings of the Royal Society of Medicine (Section of Surgery), **35,** 321.
BURNETT, C. H., SHAPIRO, S. L., SIMEONE, F. A., BEECHER, H. K., MALLORY, T. B., and SULLIVAN, E. R. (1947): Post-traumatic Renal Insufficiency. Surgery, **22,** 994.
DEROT, M., PIGNARD, P., TOURAINE, R. L., and BERNARD, J. (1953): Le potassium dans les néphropathies tubulaires anuriques. Presse Médicale, **61,** 207.
FENN, W. O. (1940): The Rôle of Potassium in Physiological Processes. Physiological Reviews, **20,** 377.
GREEN, H. N. (1945): Biochemical Factors in the Aetiology of " Shock." British Medical Bulletin, **3,** 102.

HICKS, M. H., CRUTCHFIELD, A. J., and WOOD, J. E., Jun. (1950): Intestinal Lavage in the Potassium Intoxication of Lower Nephron Nephrosis. American Journal of Medicine, **9,** 57.

HOFF, H. E., SMITH, P. K., and WINKLER, A. W. (1941): The Cause of Death in Experimental Anuria. Journal of Clinical Investigation, **20,** 607.

HUMPHREY, J. H., and AVERY JONES, F. (1947): Oliguria after Abortion. Clinical Science, **6,** 173.

KEITH, N. M., and BURCHELL, H. B. (1949): Clinical Intoxication with Potassium: its Occurrence in Severe Renal Insufficiency. American Journal of the Medical Sciences, **217,** 1.

KNOWLES, H. C., Jun., and KAPLAN, S. A. (1953): Treatment of Hyperkalemia in Acute Renal Failure using Exchange Resins. Archives of Internal Medicine, **92,** 189.

MERONEY, W. H., and HERNDON, R. F. (1954): The Management of Acute Renal Insufficiency. Journal of the American Medical Association, **155,** 877.

MUIRHEAD, E. E., and HILL, J. M. (1948): The Treatment of Acute Renal Insufficiency. Surgery, Gynecology and Obstetrics, **87,** 445.

PLATT, R. (1953): Water and Electrolytes in Renal Failure. Acta Medica Scandinavica, **146,** 39.

REWELL, R. E. (1943): Rise in Potassium Concentration in the Blood Stream following Ischaemia of Muscle Masses. British Medical Journal, **ii,** 483.

SMITH, H. W. (1951): The Kidney: Structure and Function in Health and Disease, pp. 348–349, 752–813. New York: Oxford University Press.

SMITH, L. H., Jun., POST, R. S., TESCHAN, P. E., ABERNATHY, R. S., DAVIS, J. H., GRAY, D. M., HOWARD, J. M., JOHNSON, K. E., KLOPP, E., MUNDY, R. L., O'MEARA, M. P., and RUSH, B. F., Jun. (1955): Post-traumatic Renal Insufficiency in Military Casualties: II. Management, Use of an Artificial Kidney, Prognosis. American Journal of Medicine, **18,** 187.

STOCK, R. J. (1949): Acute Urinary Suppression. Observations in Twenty-two Patients. American Journal of Medicine, **7,** 45.

STRAUSS, M. B. (1948): Acute Renal Insufficiency due to Lower Nephron Nephrosis. New England Journal of Medicine, **239,** 693.

SWANN, R. C., and MERRILL, J. P. (1953): The Clinical Course of Acute Renal Failure. Medicine, **32,** 215.

TESCHAN, P. E., POST, R. S., SMITH, L. H., Jun., ABERNATHY, R. S., DAVIS, J. H., GRAY, D. M., HOWARD, J. M., JOHNSON, K. E., KLOPP, E., MUNDY, R. L., O'MEARA, M. P., and RUSH, B. F., Jun. (1955): Post-traumatic Renal Insufficiency in Military Casualties: I. Clinical Characteristics. American Journal of Medicine, **18,** 172.

WINKLER, A. W., HOFF, H. E., and SMITH, P. K. (1941): The Toxicity of Orally Administered Potassium Salts in Renal Insufficiency. Journal of Clinical Investigation, **20,** 119.

HYPOPHOSPHATASIA

W. DICKSON, BOLTON, ENGLAND

Department of Paediatrics, Bolton Hospital Group

and

R. H. HORROCKS, BOLTON, ENGLAND

Department of Pathology, Bolton Hospital Group

In 1948 Rathbun described an infant who died at ten weeks of age with a clinical and radiological picture resembling that of rickets but who had a persistently low serum alkaline phosphatase. He named the condition hypophosphatasia. Since then further cases have been described in the literature and it appears that, although unrecognised in the past, this disease is not very uncommon.

There are varying degrees of severity of the condition. The most severe cases are usually fatal in early infancy, probably from an associated renal lesion.

FIG. 1
Right and left knees.

Milder cases may be recognised clinically from stunted growth, normal intelligence and varying degrees of bone changes of a rachitic nature. There is often a history of a rather stormy passage in early infancy, followed by some delay in reaching the normal milestones of physical development. Craniostenosis and premature loss of the deciduous teeth, not commonly associated with rickets, are sometimes found. The craniostenosis may lead to

defective vision and may necessitate surgical intervention to save sight. Looseness of the deciduous teeth may be the first feature which draws attention to the disease. The more recently investigated patients have been shown to excrete an excess of phosphoethanolamine in the urine.

A mild case of the disease is presented with some additional observations and a critical review of the literature.

CASE REPORT

A boy born in August 1946, the second child of healthy parents, weighed 5 lb. 6 oz. He did not thrive in early infancy, failed to gain weight satisfactorily and had periodic attacks of vomiting. He sat up at eight months but did not walk until he was three years of age,

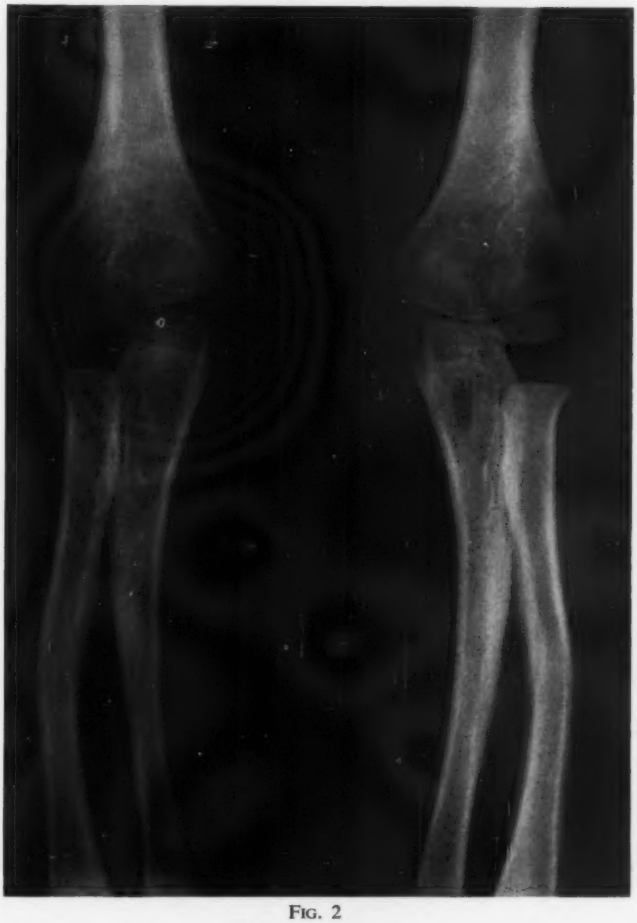

FIG. 2
Elbows and forearms.

and even then he was unsteady on his legs and fell frequently. It was not until he was five that he was able to run about like other children. His mental development seemed to be normal. He cut his first tooth at one year and had cut several by the age of two years. The teeth were poorly formed and black. They were loose, and dropped out if he banged his head. Soon after his second birthday he had lost all his teeth.

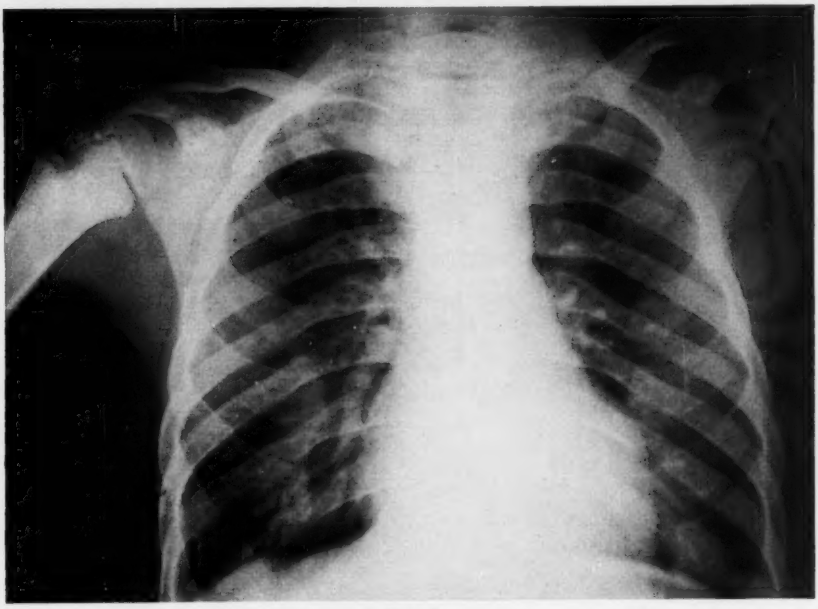

FIG. 3
Shoulders and thorax.

FIG. 4
Pelvis and hips.

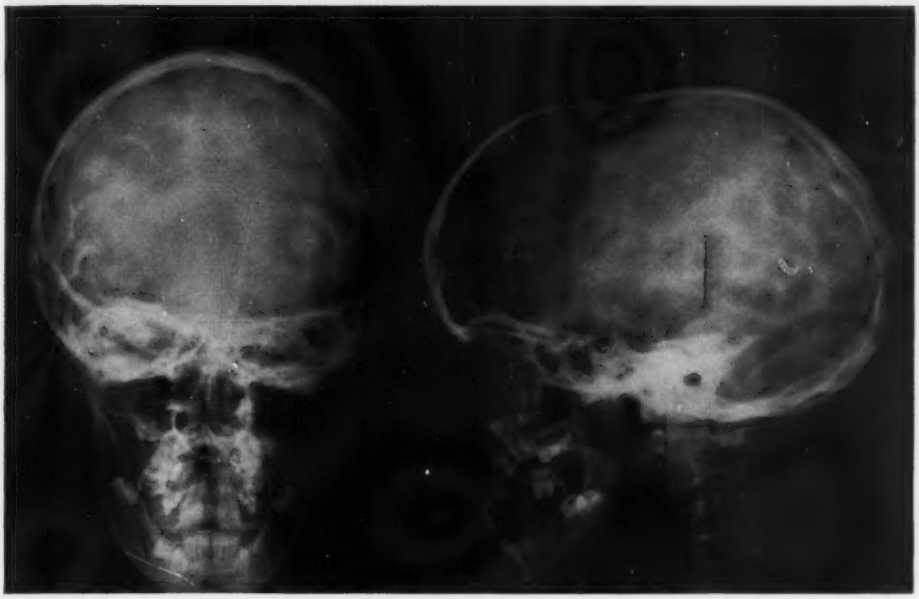

FIG. 5
Skull.

He was admitted to hospital at the age of six years with suspected acute appendicitis, but the abdominal symptoms quickly subsided. It was noted that he was small and had a peculiarly shaped head, thickening of the wrists and beading of the ribs. Radiographs of the skeleton showed changes consistent with rickets. Renal function tests and fat balance tests were normal. The serum calcium was slightly raised, the serum phosphorus normal and the urinary excretion of calcium normal. The serum alkaline phosphatase was not determined. He was considered to present a type of osteochondrodystrophy and was followed up in the out-patients' department, but after a short time failed to attend.

He was not seen again for four years when he was referred by his doctor for reassessment. He was now nearly ten years of age. His intelligence was normal; he was in a form for children of his own age; he liked school and was interested in his work. He had a peculiar shuffling gait, and if he walked any distance complained of pains in the legs and had to rest. He was slow at running, being always last in races with children of his own size but, even so, he insisted on taking part in all the sporting activities at his school. One year before, he had been admitted to another hospital with a fracture of the shaft of his left femur caused by a fall. The fracture was reduced and immobilised in a plaster spica, and united soundly within two months.

On examination he was a small, normally proportioned child three feet eight inches in height (eight inches below average). He had a long scaphocephalic type of skull and a somewhat depressed bridge of nose. He had no deciduous teeth and only four permanent teeth, which were normal. All the first permanent molars had been extracted for caries. His eyes were brown. He was short-sighted and wore glasses. The optic discs were normal. The bone deformities observed in 1952 were still present. Further radiographs of the skeleton showed changes like those of 1952 and they are now described in detail.

Radiological features—The skeleton was poorly mineralised and the changes were most marked at the metaphysial region of the long bones and at the sites of most active growth, namely the

knee, shoulder and wrist joints. *Knee joints* (Fig. 1): Here the typical changes were most clearly seen. The epiphyses were poorly calcified. The metaphysial line was irregular. There was some trumpeting of the lower end of both femurs, but no cupping. The femoral metaphyses were irregularly calcified and resembled the end of a " rotten stump." The upper ends of the

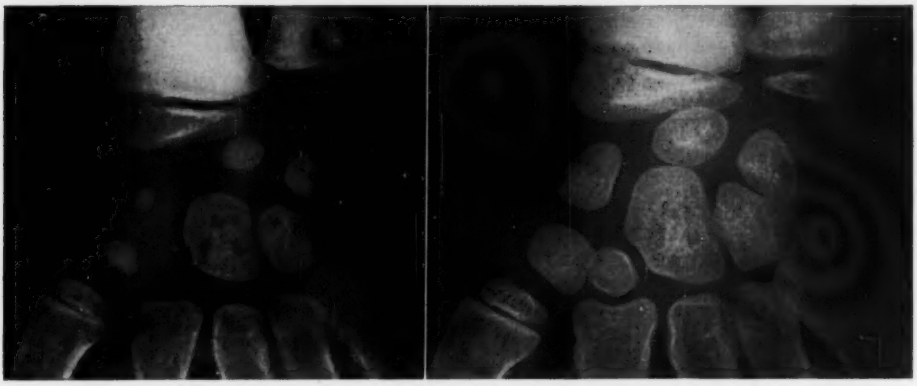

FIG. 6 FIG. 7

Wrist. Figure 6—In 1952. Figure 7—In 1956.

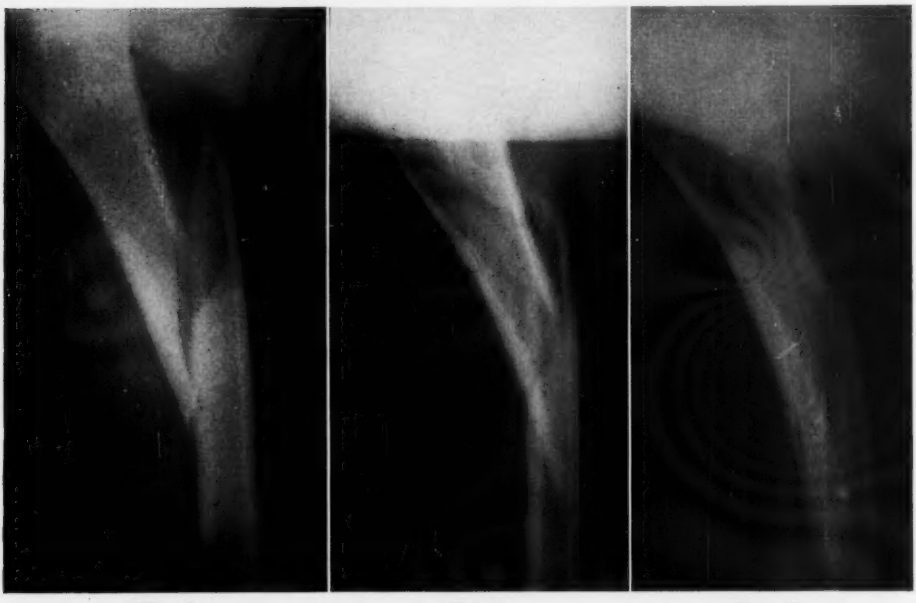

FIG. 8 FIG. 9 FIG. 10

Fracture of femur 1955. Figure 8—Original fracture. Figure 9—Two months later. Figure 10—One year later.

fibulae had a cystic appearance. The left femoral metaphysis was more severely affected than the right for reasons which will be discussed later. *Elbow joints and forearms* (Fig. 2): The elbows showed similar changes, in milder degree. There was outward bowing of the radius and ulna. *Shoulder joints and thorax* (Fig. 3): The scapulae were poorly mineralised and the

distal ends of the ribs expanded. The upper end of the right humerus was clearly affected. *Pelvis and hip joints* (Fig. 4): The proximal metaphyses of the femurs (particularly the left) showed distinct changes. The bones of the pelvis and spine were poorly mineralised; in the spine mineralisation improved from below upwards, reaching the normal in the cervical region. The iliac bones were grossly demineralised. *Skull* (Fig. 5): The sagittal suture had fused prematurely producing a long scaphocephalic type of skull. The bones of the vault were thinner than normal and there was a beaten silver appearance. The teeth were deficient: there were no deciduous teeth; only four permanent teeth had erupted and several permanent teeth had failed to form. *Wrists* (Figs. 6 and 7): The bone age was only slightly retarded and the development during the previous four years was normal. *Fracture of the femur* (1955): The fracture was shown to have united readily (Figs. 8 to 10).

TABLE I

SEROLOGICAL INVESTIGATIONS

Date	Alkaline phosphatase (*K.-A. units*)	Calcium (*mg./100 ml.*)	Inorganic phosphorus (*mg./100 ml.*)	Urea (*mg./100 ml.*)	Cholesterol (*mg./100 ml.*)	Sodium (*mEq./litre*)	Potassium (*mEq./litre*)	Chloride (*mEq./litre*)	Alkali reserve (*mEq./litre*)
12.11.52	—	12·5	5·8	26	—	—	—	—	—
4.2.56	3·4	10·2	5·8	29	180	—	—	—	—
5.3.56	2·0	9·9	6·5	30	—	138	—	108	—
12.3.56	1·5	10·2	6·2	41	—	135	4·1	103	18
6.4.56	4·2	10·3	—	33	170	143	4·0	96	—
18.4.56	2·7	10·5	6·0	18	—	—	—	—	—
23.4.56	2·1	11·4	—	—	—	—	—	—	—
25.4.46	3·0	10·3	5·0	—	180	—	—	—	—
3.5.56	2·8	12·0	—	24	—	—	—	—	—
10.5.56	2·8	11·3	—	29	—	—	—	—	—
17.5.56	3·1	11·2	—	22	—	—	—	—	—

Investigations—The serum chemistry results, during four months, are shown in Table I. Calcium, inorganic phosphorus and alkaline phosphatase are recorded graphically in Figure 11. The serum alkaline phosphatase was persistently low. Serum inorganic phosphorus was slightly raised on most occasions. Serum calcium levels were within normal limits except on two occasions: in 1952 when a value of 12·5 milligrams per cent was recorded, and after vitamin D therapy in 1956 when a level of 12·0 milligrams per cent was reached.

Urinary calcium and phosphorus excretion was within normal limits. Renal function was normal as judged by serum urea estimations, urine concentration and urea clearance tests. Excretion urography showed the kidneys to be normal in size and shape and well able to concentrate the dye. Examination of the stools for fat on several occasions showed no evidence of steatorrhoea. Wassermann and Kahn tests were negative. The glucose tolerance and vitamin C saturation tests were normal. A considerable quantity of phosphoethanolamine was found in the urine (for this investigation we are indebted to Professor C. E. Dent and Dr H. Harris).

Heredity—Dr H. Harris has not yet completed the family survey with regard to phosphoethanolamine, but he reports that he has found significant amounts of phosphoethanolamine

in the urine of the two brothers, both parents, and an aunt and uncle on both paternal and maternal sides of the family. Both parents and the two siblings are below average height. Radiographs of their skeletons show no abnormality. There is a tendency to low serum alkaline phosphatase in the mother and two brothers: mother (aged thirty-three) 3·7 and 5·5 King-Armstrong units; brother (aged thirteen) 9·7 King-Armstrong units; brother (aged five) 8·0 King-Armstrong units.

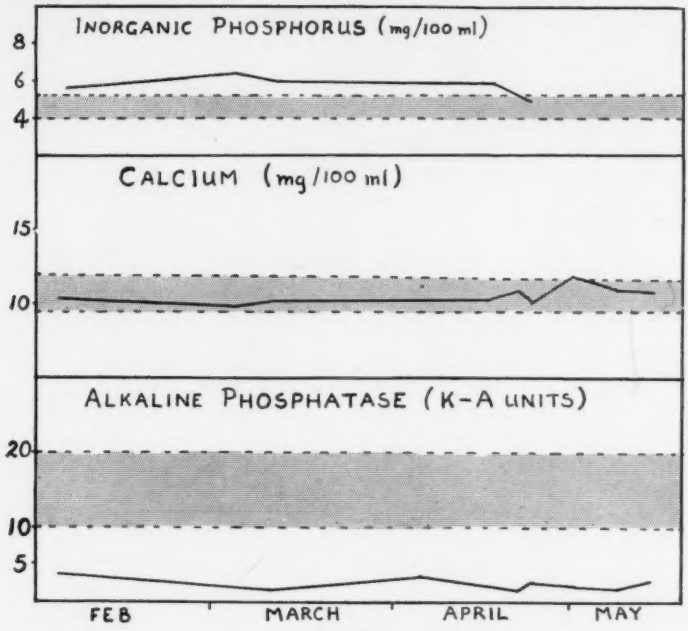

FIG. 11

Serum alkaline phosphatase, calcium and inorganic phosphorus during four months. The shaded areas represent the normal range in each case.

REVIEW OF THE LITERATURE

Twenty-two cases in the literature have been reviewed and it appears that hypophosphatasia may be divided into three groups. 1) Severe cases, fatal in the first year of life. 2) Moderately severe cases usually presenting later in childhood, in which the condition remains stationary or improves. 3) Mild cases in patients who may have no bony changes but alkaline phosphatase on the low side of normal and excessive excretion of phosphoethanolamine in the urine.

The findings in seventeen cases are summarised in Tables II and III.

Group 1 (Table II)—In the six fatal cases the patients all died within the first year of life. Four of these cases have been fully reported in the literature (Rathbun 1948, Engfeldt and Zetterström 1954, 1956; McCance *et al.* 1956). None of the patients made satisfactory progress from birth; they failed to gain weight, had periodic attacks of vomiting and intermittent cyanosis. The serum alkaline phosphatase level was lower than in the other groups, and in Rathbun's case alkaline phosphatase was almost absent from the serum. Alkaline phosphatase has been shown to be markedly deficient in all organs where it is normally present.

Varying degrees of hypercalcaemia and hyperphosphataemia were present in all cases in which these investigations were carried out.

All children had gross skeletal deformities, radiographic changes resembling those of severe rickets, and fractures of the ribs and of the metaphysial regions of the long bones.

TABLE II

TABLE II

REPORTED FATAL CASES OF HYPOPHOSPHATASIA

Author	Age at death	Sex	Physical deformities of severe rickets	Radiological appearances resembling rickets	Skull, soft like leather	Fontanelle bulging	Fractures	Shortening of limbs	Renal damage	Serum alkaline phosphatase	Serum calcium
Rathbun (1948)	10 weeks	M	Yes	Yes	Yes	Not reported	Yes	Yes	Yes	Very low	Raised
Sobel *et al.* (1953)	8 months	Not stated	Yes	Yes	Not reported	Not reported	Not reported	Not reported	Yes	Low	Not reported
Sobel *et al.* (1953)	8 months	Not stated	Yes	Yes	Not reported	Not reported	Not reported	Not reported	Yes	Low	Not reported
Engfeldt and Zetterström (1954)	10 months	F	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Low	Raised
McCance *et al.* (1956)	14 weeks	F	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Low in tissues	Raised
McCance *et al.* (1956)	16 weeks	M	Yes	Yes	Yes	Not reported	Yes	Yes	Yes	Low	Raised

TABLE III

REPORTED MODERATE AND MILD CASES OF HYPOPHOSPHATASIA

Author	Age	Sex	Rachitic deformities	Craniostenosis	Premature loss of milk teeth	Renal damage	Serum alkaline phosphatase	Serum calcium	Serum inorganic phosphorus
Schneider and Corcoran (1950)	10 years	M	Yes	Not reported	Not reported	Excessive phosphate reabsorption	Low	Raised	Raised
Schneider and Corcoran (1950)	13 years	M	Yes	Not reported	Not reported	Excessive phosphate reabsorption	Low	Raised	Raised
Sobel *et al.* (1953)	19 months	F	Yes	No	Yes	None apparent	Low	Normal	Normal
Sobel *et al.* (1953)	20 months	M	Yes	Not reported	Yes	None apparent	Low	Not reported	Not reported
Sobel *et al.* (1953)	6 months	F	Yes	Not reported	Yes	Not reported	Low	Not reported	Not reported
Sobel *et al.* (1953)	8½ years	Not stated	Yes	Not reported	Yes	Not reported	Low	Not reported	Not reported
Schlesinger *et al.* (1955)	7½ months	F	Yes	Yes	No	Yes	Low	Raised	Low
Schlesinger *et al.* (1955)	6 months	F	Yes	Yes	No	None apparent	Low	Normal	Normal
Schlesinger *et al.* (1955)	2½ years	F	Yes	Yes	No	None apparent	Low	Raised	Not reported
Illingworth and Gardiner (1955)	4 years	M	Bowing of legs	No	Yes	None apparent	Low	Normal	Normal
Fraser *et al.* (1955)	3 months	F	Yes	No		Mild insufficiency	Low	Not reported	Not reported

Old healed fractures were demonstrated at necropsy. A soft leather-like skull was a common feature. Deaths have been attributed to renal tubular lesions secondary to hypercalcaemia.

Group 2 (Table III)—Of the eleven patients reviewed, only four were under the age of one year when the disease was diagnosed. A history of a stormy passage in the first six months of life was remarked upon in some of the cases. The alkaline phosphatase was low in all cases in the serum and also in other tissues where biopsies were taken. Of seven patients in whom the serum calcium was estimated four had a moderate hypercalcaemia. Two out of six patients had a mild hyperphosphataemia. Clinically and radiologically most of these cases resemble rickets but may have two features not commonly associated with rickets, namely craniostenosis and premature loss of the deciduous teeth. An increased liability to fracture was not noted in the group. Only three patients showed evidence of impaired renal function.

Group 3—In these cases there appear to be no symptoms, and as a rule they are discovered as a result of a family survey of affected children. Dent (1956) referred to an adult who had phosphoethanolamine in the urine and an increased liability to fractures.

DISCUSSION

Fatal cases—From the cases reported so far it seems that the critical period is the first twelve months of life. If an infant can survive this period without any serious renal impairment his chances of survival are good, and there is every possibility that the condition will improve or remain stationary. In the fatal cases the serum alkaline phosphatase was generally lower than in the non-fatal cases, but this is of doubtful prognostic value because in the case described, and in some previously reported cases, very low levels were reported in the milder forms.

The cause of death in these fatal cases has been attributed to renal damage secondary to hypercalcaemia. However, in idiopathic hypercalcaemia, for instance, the serum calcium often reaches much higher levels without any serious or permanent renal damage.

The reason for the renal tubular lesions and the hypercalcaemia is obscure, but it is likely that the hypophosphatasia is responsible. Although the clinical results of the phosphatase deficiency are mainly manifested in the skeleton, it must be remembered that there is a generalised tissue deficiency of alkaline phosphatase in this disease.

The results of this deficiency in other organs, including the kidneys, may contribute to the cause of death.

Skeletal growth—It seems that in the patients who survive infancy the need for alkaline phosphatase grows less and that the severity of the bone changes depends on the rate of growth and the extent of the enzyme deficiency. In the bones that develop from cartilage, growth in length is impaired. In the case reported here, and in the affected siblings, the long bones were much shorter than normal and growth appeared to be affected more than in vitamin D deficiency rickets. It seems that, in the milder cases, as the rate of growth slows down, the bone changes improve or remain stationary and in some there is sufficient alkaline phosphatase for them to return to normal (Schlesinger *et al.* 1955 (Case 2)).

If the rate of growth increases the bone changes worsen. Overgrowth of a bone occasionally follows the fracture of the shaft of a long bone during the period of growth. Trueta (1953) found, in rabbits, that if the medullary cavity of the shaft of a long bone was blocked, with consequent interference with the circulation, increased growth occurred at the epiphysial plate. He attributed this increased growth to an increase in the blood supply of the metaphysis from the collateral circulation through vessels entering the bone in the metaphysial region. A similar change occurred in this case (Fig. 7). After the fracture of the mid-shaft of the left femur an overgrowth of this bone has occurred. This has caused a worsening of the condition in the metaphysial regions of both ends of the bone. In 1952 the changes in both femurs were the same but, in the radiographs taken in 1956, a year after the fracture, the changes are more marked in the fractured bone. The deterioration in this bone can only be attributed to its increased rate of growth.

Deformities—In early life the bones appear to be softer than normal and liable to the same deformities as the bones in rickets, especially the weight-bearing bones. Our patient was late in walking and for a long time crawled about on all fours; so weight bearing on his legs was delayed until the bones were harder, thus preventing deformity of the legs. Nevertheless there is now outward bowing of both forearms, and this deformity seems to have been produced by the prolonged period of crawling. Many of these children were slow in reaching the physical milestones of development, which protected them from bony deformity. It would probably be advisable in mild cases to delay weight bearing until the condition improves.

Fractures—In many of the severe cases fractures through the metaphysial regions of the long bones have been reported. There is a definite weakness in this region in the young child. Dent (1956) suggested that, in later life, some cases may present with spontaneous fractures. There is no published evidence of increased liability to fracture in older children. Our patient did sustain a fracture of the mid-shaft of the femur, but the injury was severe and probably would have caused a fracture in a normal child.

It is known that alkaline phosphatase plays an important part in the healing of fractures. At a fracture site marked phosphatase activity has been noted experimentally. Nevertheless the fracture in our patient healed without delay in spite of the marked deficiency of alkaline phosphatase in the serum. It is also interesting that in some of the severe cases, with almost total absence of alkaline phosphatase, healed fractures were seen. On the other hand Schlesinger *et al.* (1955) reported delay in healing of a parietal burr hole, and Schneider and Corcoran (1950) found delay in the healing of osteotomies performed to correct deformities. Nevertheless it seems that in most cases there is sufficient alkaline phosphatase present, or sufficient is able to be produced locally, to enable normal repair of the fracture to occur.

Craniostenosis—In the skull growth of bone is affected, and as the children grow older they are liable to develop craniostenosis. Schlesinger *et al.* (1955) first drew attention to this complication, and they also pointed out that it has been reported in cases of vitamin D resistant rickets (Coleman and Foote 1954). They suggested that cessation of normal growth at the suture line is the decisive factor. In our own case the sagittal suture was involved.

Premature loss of deciduous teeth—Premature loss of the deciduous teeth was first reported by Sobel *et al.* (1953) and later by Illingworth and Gardiner (1955), whose patient had incomplete formation and premature absorption of the roots of some exfoliated teeth. Our patient had lost his deciduous teeth when first examined, but the roots of two of the sixth-year molars, extracted on account of caries, were normal. We have noted looseness of the deciduous teeth in two brothers with a rare type of rickets, and it is probably an osteomalacic manifestation. The failure of some permanent teeth to form in our case is difficult to explain and is not regarded as part of the clinical picture of the disease.

Comments—The etiology of hypophosphatasia is unknown, but the cases reported suggest that the abnormality is genetically determined.

The exact function of alkaline phosphatase in bone metabolism is still obscure. According to Bourne (1956) phosphatase is thought to be more concerned with the formation of the organic matrix of bone rather than with the precipitation or crystallisation of bone salts. Rathbun (1948) and Sobel *et al.* (1953) both showed that not only the skeleton but other tissues, normally rich in phosphatase, may be grossly deficient in this enzyme, which suggests a general cellular dysfunction or defect in body synthesis of alkaline phosphatase. Rathbun (1948) and Schlesinger *et al.* (1955) suggested that there may be a primary defect in the development of osteoblasts resulting in an insufficiency of alkaline phosphatase. However, in other reported cases the osteoblasts did not appear to be deficient either in quality or quantity on histological examination, and other functions of the osteoblasts, such as repair of fracture, were carried out normally. The low serum alkaline phosphatase could be due to the presence of an inhibitor, but it has been shown by Fraser *et al.* (1955), Sobel *et al.* (1953) and Engfeldt and Zetterström (1954, 1956) that serum from such cases does not inhibit the

alkaline phosphatase activity of normal serum. Fraser *et al.* (1955) showed that serum from a patient with hypophosphatasia will produce *in vivo* calcification in the cartilage of rachitic rats and concluded that defective mineralisation is due to a primary disturbance in the metabolism of the bone matrix.

The discovery of phosphoethanolamine in the urine and plasma of later cases (Fraser *et al.* 1955, McCance *et al.* 1955, 1956; Dent and Harris 1956) may shed further light on the function of alkaline phosphatase in bone metabolism. As Dent (1956) suggested, it may be that phosphoethanolamine is one of the substrates for bone alkaline phosphatase and that in hypophosphatasia there is a metabolic block resulting from an inborn error of metabolism.

SUMMARY

1. A case of hypophosphatasia in a boy who sustained a fractured left femur is described.
2. The literature is reviewed, and the reported cases are found to fall into severe, moderately severe and mild forms.
3. The diagnostic features of the disease are the radiological picture, which resembles that of rickets, very low serum alkaline phosphatase, and excessive phosphoethanolamine excretion in the urine.
4. Other clinical features may be a failure to thrive in early infancy, premature loss of deciduous teeth, hypercalcaemia and renal damage.
5. The function of alkaline phosphatase in bone metabolism in relation to this disease has been discussed.

We wish to thank Dr S. W. Stanbury, Dr G. B. Manning, Dr D. M. Barry, Dr J. R. Kirby, Mr F. Taylor Monks and Dr S. A. Narayan for their help in preparing this paper. We also thank Mr Kilshaw for the clinical photographs and Dr D. J. Gracie for referring the patient to us.

REFERENCES

BOURNE, G. H., ed. (1956): The Biochemistry and Physiology of Bone, Chapter 9. London: Academic Books Ltd.
COLEMAN, E. N., and FOOTE, J. B. (1954): Craniostenosis with Familial Vitamin-D-resistant Rickets. British Medical Journal, **i,** 561.
DENT, C. E. (1956): *In* Ciba Foundation Symposium on Bone Structure and Metabolism, p. 267. Edited by G. E. W. Wolstenholme and C. M. O'Connor. London: J. & A. Churchill Ltd.
DENT, C. E., and HARRIS, H. (1956): Hereditary Forms of Rickets and Osteomalacia. Journal of Bone and Joint Surgery, **38–B,** 204.
ENGFELDT, B., and ZETTERSTRÖM, R. (1954): Osteodysmetamorphosis Fetalis. Journal of Pediatrics, **45,** 125.
ENGFELDT, B., and ZETTERSTRÖM, R. (1956): Osteodysmetamorphosis Foetalis. *In* Ciba Foundation Symposium on Bone Structure and Metabolism, edited by G. E. W. Wolstenholme and C. M. O'Connor, p. 258. London: J. & A. Churchill Ltd.
FRASER, D., YENDT, E. R., and CHRISTIE, F. H. E. (1955): Metabolic Abnormalities in Hypophosphatasia. Lancet, **i,** 286.
ILLINGWORTH, R. S., and GARDINER, J. H. (1955): Premature Loss of Deciduous Teeth. Archives of Disease in Childhood, **30,** 449.
LANMAN, J. T. (1955): Hypophosphatasia—A Newly Recognized Disease. Journal of Pediatrics, **47,** 509.
MCCANCE, R. A., MORRISON, A. B., and DENT, C. E. (1955): The Excretion of Phosphoethanolamine and Hypophosphatasia. Lancet, **i,** 131.
MCCANCE, R. A., FAIRWEATHER, D. V. I., BARRETT, A. M., and MORRISON, A. B. (1956): Genetic, Clinical, Biochemical, and Pathological Features of Hypophosphatasia. Quarterly Journal of Medicine, **25,** 523.
RATHBUN, J. C. (1948): " Hypophosphatasia." American Journal of Diseases of Children, **75,** 822.
SCHLESINGER, B., LUDER, J., and BODIAN, M. (1955): Rickets with Alkaline Phosphatase Deficiency. An Osteoblastic Dysplasia. Archives of Disease in Childhood, **30,** 265.
SCHNEIDER, R. N., and CORCORAN, A. C. (1950): Familial Nephrogenic Osteopathy due to Excessive Tubular Reabsorption of Inorganic Phosphate. Journal of Laboratory and Clinical Medicine, **36,** 985.
SOBEL, E. H., CLARK, L. C., Jun., FOX, R. P., and ROBINOW, M. (1953): Rickets. Deficiency of " Alkaline " Phosphatase Activity and Premature Loss of Teeth in Childhood. Pediatrics, **11,** 309.
TRUETA, J. (1953): The Influence of the Blood Supply in Controlling Bone Growth. Bulletin of the Hospital for Joint Diseases, **14,** 147.

THE RESULTS OF PARTIAL AND TOTAL EXCISION OF THE PATELLA

H. L. DUTHIE, GLASGOW, SCOTLAND, and J. R. HUTCHINSON, BURNLEY, ENGLAND

From the Western Infirmary, Glasgow

Opinions differ on the effects of excision of the patella. Good results were reported by Blodgett and Fairchild (1936), by Dobbie and Ryerson (1942) and by Horowitz and Lambert (1946). Indeed Brooke (1937) stated that the function of the knee was better than the normal after removal of the patella, and that the patella was not morphologically an integral part of the quadriceps tendon.

These opinions are in conflict with those of Haxton (1945) and Jensenius (1951) who showed that extension of the knee was impaired by removing the patella. Bruce and Walmsley (1942) demonstrated early and severe arthritic changes in the femoral condyles of rabbits when the patella had been removed. Cohn (1944) showed that these changes were reduced when part of the patella was retained, and recommended partial excision whenever possible. Thomson (1942) recorded good results from this procedure in a follow-up by questionnaire.

TABLE I

ANALYSIS OF PATIENTS EXAMINED

Total excision of patella ..	56
For chondromalacia	32
For fracture	24
Partial excision of patella .	21

TABLE II

DURATION OF FOLLOW-UP

	1–2 years	3–5 years	6–10 years
Total excision of patella			
For chondromalacia	9	18	5
For fracture . .	9	8	7
Partial excision of patella	7	12	2

Since there is no firm opinion of the merits of complete, as distinct from partial, excision, it was decided to review the results of the two operations, as practised in one unit during the past ten years.

MATERIAL

In the ten years under review the patella was excised completely or in part in 146 patients. Seventy-seven of these were examined (Table I).

The remaining sixty-nine patients were not seen for the following reasons: seven had died; thirty-six had moved and could not be traced; ten reported satisfactory results by letter but were too far away to attend; and sixteen failed to respond.

The follow-up was from one to ten years after operation (Table II).

All the patients with fractures were operated upon within two weeks of the accident, except one in whom other injuries caused a delay of two months. The following operations were performed.

Partial excision for fracture—The proximal pole was retained and the ligamentum patellae were reattached to the bone (Thomson 1942). The lateral expansions of the quadriceps were repaired.

Total excision for fracture—The patella was excised. In approximately half the patients the quadriceps tendon was sutured to the ligamentum patellae, and in the remainder an overlapping repair of the aponeurosis was done. These variations of technique did not affect the ultimate result.

Total excision for chondromalacia—The patella was excised through a transverse incision in the aponeurosis in eighteen patients, and through a vertical incision in the remaining fourteen.

In twenty-four patients the knee was splinted in a plaster cylinder for two to four weeks after the operation; in the remainder only a compression bandage was used. Quadriceps contractions were begun the day after operation; knee flexion was allowed after two weeks when the patella was removed through a vertical incision and after three to four weeks in the rest. Weight bearing was allowed when the knee could be flexed to a right angle under good muscle control. The type or duration of splintage did not affect the results.

Complications were recorded in seven patients: haematoma (four cases); haemarthrosis requiring aspiration (two cases); rupture of the suture line necessitating further repair (one case).

METHOD OF ASSESSMENT

Each patient was interviewed and examined, and radiographs were taken. Note was made of the development of the quadriceps muscle, and the quadriceps power in fullest extension of the knee was determined against a spring balance (Fig. 1). An average of three readings was taken and expressed as a percentage of the power in the normal knee.

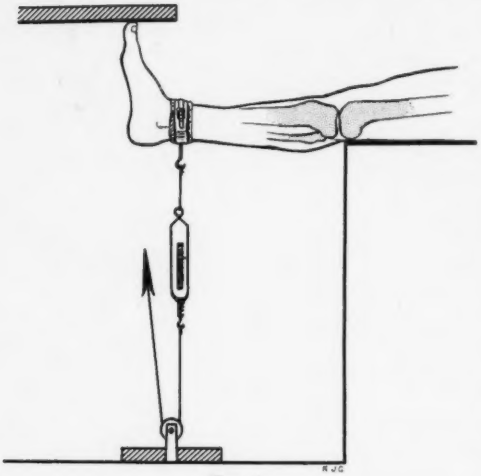

FIG. 1

Apparatus used for estimating quadriceps power in maximum extension. One observer applies the force represented by the arrow and notes when the big toe is pulled away from the mark. The other observer records the tension as shown on the spring balance.

The results were graded by the following subjective and objective criteria. In cases classed as *excellent* the knee was indistinguishable functionally from normal. With a *good* result there was minor stiffness or aching after prolonged sitting, slight subjective weakness, slight tenderness, or wasting by less than half an inch. Power was at least 90 per cent of the normal. With a *fair* result there were pain and stiffness after sitting, weakness on stairs, sufficient tenderness to make kneeling uncomfortable, loss of flexion up to 30 degrees, or loss of extension up to 5 degrees. Power was not less than 75 per cent of normal. The result was classed as *bad* in all patients who failed to attain the above standards. The function of the knee was judged to have been seriously affected, although the patient may have returned to his former employment.

RESULTS

The results in the seventy-seven patients are shown in Table III.

In many cases considerable time elapsed after the operation before the patient considered that the knee had ceased to improve. The time taken for greatest recovery is detailed in Table IV.

The main symptoms and clinical findings at follow-up are listed in Table V.

DISCUSSION

Most patients do not recover greatest function in the knee until between six and twelve months after operation—a longer delay than has usually been reported. The better results

TABLE III

RESULTS OF SEVENTY-SEVEN OPERATIONS FOR TOTAL OR PARTIAL EXCISION OF THE PATELLA

	Result			
	Excellent	Good	Fair	Bad
Total excision of patella				
For chondromalacia (32 cases)	5	16	7	4
For fracture (24 cases) . .	2	10	4	8
Partial excision of patella (21 cases)	3	6	5	7

TABLE IV

TIME TAKEN FOR FULLEST RECOVERY

	Time to greatest recovery			
	1–3 months	4–6 months	7–12 months	13–18 months
Total excision of patella				
For chondromalacia (32 cases)	5	4	14	9
For fracture (24 cases) . .	3	2	15	4
Partial excision of patella (21 cases)	5	4	11	1

TABLE V

MAIN SYMPTOMS AND CLINICAL FINDINGS AT TIME OF FOLLOW-UP EXAMINATION

	Total excision of patella		Partial excision of patella (21 cases)
	For chondromalacia (32 cases)	For fracture (24 cases)	
Aching at rest	8	6	7
Pain	8	9	7
Subjective weakness . . .	15	19	14
Stiffness after rest . . .	16	14	15
Loss of full extension . .	4	5	4
Loss of flexion			
10–30 degrees . . .	—	2	4
Over 30 degrees . . .	4	6	4
Wasting of thigh			
½–1 inch	11	3	6
Over 1 inch . . .	5	10	7
Quadriceps power			
75–90% of sound side. .	8	4	5
Less than 75% of sound side	1	8	10
Degeneration of femoral condyle			
Moderate	4	10	12
Severe	2	2	3
Ossification in quadriceps tendon			
Slight	10	8	2
Severe	3	6	—
Change of occupation . .	3	3	3

in the subjects of chondromalacia than in those with fracture, however treated, may be explained partly by the younger age of the patients (average 25·6 years as against 47·7 years). After complete excision of the patella, subjective weakness was present more often than demonstrable wasting or loss of power of the quadriceps muscle (Fig. 2). Jensenius (1951) suggested that weakening of knee extension after excision of the patella is due to shortening

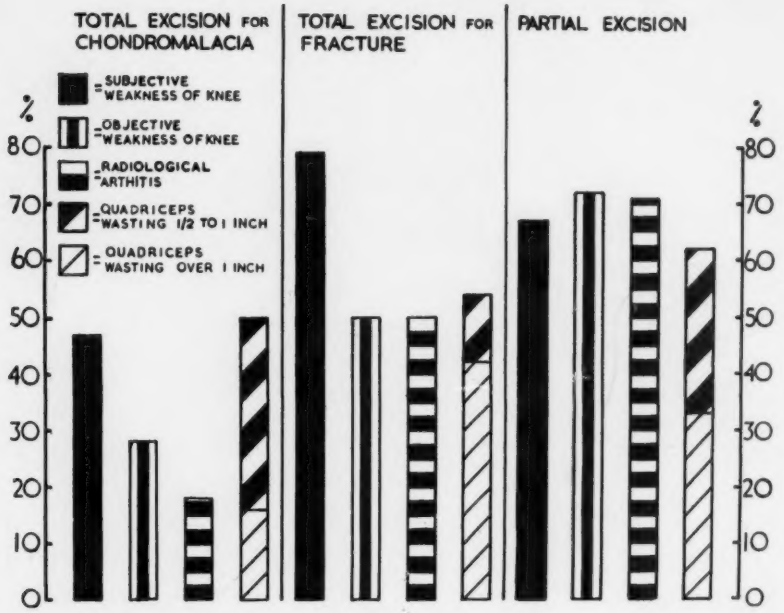

FIG. 2
Comparison of subjective and objective findings expressed as percentage of group totals.

of the distance between the axis of movement of the knee and the quadriceps tendon. Were this the case one would expect subjective and objective weakness. The feeling of weakness found in so many patients has not been satisfactorily explained. This symptom, which was in many cases trivial, could not be related to operative technique or clinical findings. However, when only part of the patella is removed, weakness and loss of power are related to the presence of radiological arthritis (Fig. 2).

To clarify further discussion the fair and bad results have been analysed together under the heading of poor results (Table VI).

The average age of patients with poor results in all groups was 45·4 years compared with 38·9 years in those with good results and 28·3 years in those with excellent results.

Most of the patients with poor results had ossification in the quadriceps tendon, and this was marked in about two-thirds of these patients (Fig. 3). Ossification of similar degree was found in only 6 per cent of those with good results. It seems likely, therefore, that discomfort in the knee has a direct relationship to ossification in the quadriceps tendon.

It has been suggested that regeneration of bone in the patellar bed is an advantage, and " subperiosteal " excision of the patella has been advised (Dobbie and Ryerson 1942). In the series there were two cases with regeneration of the patella (Fig. 4), both with poor results.

The greater incidence of arthritis of the femoral condyle in the cases of fracture may be due partly to the initial injury. We could find no evidence that excision of the patella itself

FIG. 3

FIG. 4

Figure 3—Radiograph showing ossification in the quadriceps tendon after total excision of the patella.
Figure 4—Radiograph showing regeneration of the patella after total excision.

FIG. 5

FIG. 6

Figure 5—Tilting of the patellar remnant towards the femoral condyles after partial excision of the bone.
Figure 6—Radiograph showing good alignment of the patellar remnant.

produced arthritis, as in animals. Nevertheless a significant cause of patello-femoral arthritis was evident in the cases of partial excision of the patella, for, when gross arthritic changes occurred, the lower pole of the patellar remnant was tilted towards the articular surface of the femur. This tilting was present in five out of seven patients with arthritis (Figs. 5 and 6).

TABLE VI

ANALYSIS OF FAIR AND BAD RESULTS

	Total excision of patella		Partial excision of patella (12 cases)
	For chondromalacia (11 cases)	For fracture (12 cases)	
Age over 40	4	7	10
Ossification in quadriceps .	8	10	2
Femoral arthritis . . .	4	8	11
Wasting of thigh over ½ inch .	5	9	8
Obvious weakness over 10% .	8	8	10

This malalignment is due to reattachment of the ligamentum patellae to the front of the remaining fragment of the patella. It is therefore suggested that the ligament should be sutured to the bone as near to the articular surface as possible (Fig. 7).

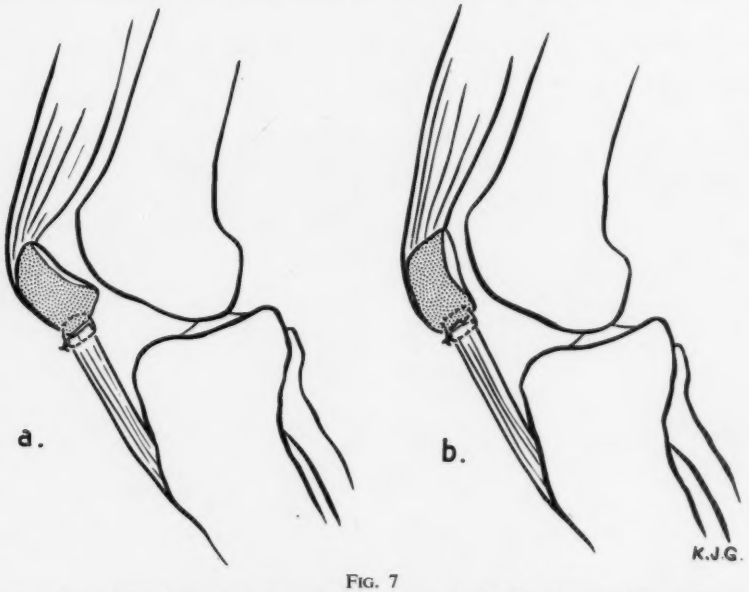

FIG. 7

Attachment of patellar tendon after partial excision giving (*left*) tilting of patellar remnant, and (*right*) good alignment.

No disadvantage has been found in excising the intact patella through a vertical split in the aponeurosis, in spite of the suggestion that this leads to loss of full extension of the knee (Smillie 1951). Among the fourteen patients in whom the patella was excised through a vertical split there was only one who could not extend the knee fully.

CONCLUSIONS

The results of our series fall between the extremes of some previous reports. The treatment of fracture of the patella is still a difficult problem, and neither of the procedures reviewed is much superior to the other. As arthritic symptoms were the main cause of disability after partial excision of the patella, it is suggested that when there is already evidence of patello-femoral arthritis before the operation it is preferable to excise the whole bone. In the younger patients partial excision may be expected to give a good result when care is taken to reattach the ligamentum patellae to the correct site.

SUMMARY

1. Seventy-seven operations on the patella have been reviewed.
2. There is no evidence that arthritic changes in the femoral condyles are an inevitable sequel of complete excision of the patella.
3. There is a direct relationship between the severity of symptoms after complete excision of the patella and the extent of ossification in the quadriceps tendon.
4. Patello-femoral arthritis after partial excision of the patella may be due to faulty realignment of the patellar ligament and consequent tilting of the patellar remnant towards the femoral condyles.

We would like to thank Mr Roland Barnes for permission to use his cases and for his help and advice in the preparation of this paper. We would also like to thank the staff of the Department of Medical Illustration of the Western Infirmary for the illustrations.

REFERENCES

BLODGETT, W. E., and FAIRCHILD, R. D. (1936): Fractures of the Patella. Journal of the American Medical Association, **106,** 2.121.

BROOKE, R. (1937): The Treatment of Fractured Patella by Excision. British Journal of Surgery, **24,** 733.

BRUCE, J., and WALMSLEY, R. (1942): Excision of the Patella. Some Experimental and Anatomical Observations. Journal of Bone and Joint Surgery, **24,** 311.

COHN, B. N. E. (1944): Total and Partial Patellectomy. An Experimental Study. Surgery, Gynecology and Obstetrics, **79,** 526.

DOBBIE, R. P., and RYERSON, S. (1942): The Treatment of Fractured Patella by Excision. American Journal of Surgery, N.S. **55,** 339.

HAXTON, H. (1945): The Function of the Patella and the Effects of its Excision. Surgery, Gynecology and Obstetrics, **80,** 389.

HOROWITZ, T., and LAMBERT, R. G. (1946): Patellectomy in the Military Service. Surgery, Gynecology and Obstetrics, **82,** 423.

JENSENIUS, H. (1951): On the Result of Excision of the Fractured Patella. Acta Chirurgica Scandinavica, **102,** 275.

SMILLIE, I. S. (1951): Injuries of the Knee Joint. Second edition, p. 230. Edinburgh: E. & S. Livingstone Ltd.

THOMSON, J. E. M. (1942): Fracture of the Patella Treated by Removal of the Loose Fragments and Plastic Repair of the Tendon. Surgery, Gynecology and Obstetrics, **74,** 860.

AN UNUSUAL MALFORMATION OF THE RADIUS AND ULNA

R. G. HARRISON and E. N. WARDLE, LIVERPOOL, ENGLAND

From the Departments of Anatomy and Orthopaedics, University of Liverpool

A girl aged eight years had noticed deformity and swelling at the lower end of the forearm for one year. She did not complain, but her mother was concerned because the arm was " growing more crooked." There was no history of injury. The child had been normal from birth, and she had suffered no serious illness.

On examination, the left forearm had an ill defined, bony-hard swelling on the extensor aspect in the lower third with slight ulnar deviation at the wrist. Function was normal at the elbow, wrist and fingers. There was no evidence of a nerve or vascular lesion.

FIG. 1

FIG. 2

Figure 1—Radiograph of the forearm. The white line, indicating the axis about which pronation and supination occur, passes through the false joint. Figure 2—Radiograph two years after excision of the exostoses.

Radiographic examination—An irregular outgrowth from the ulnar border of the radius terminated in a knob of bone which fitted into a hollow moulded in an eminence of bone arising from the radial border of the ulna (Fig. 1). The ulnar eminence was based upon an area of compact bone, much thicker than that seen elsewhere in the shaft.

Operative findings—Removal of these bony processes was advised to prevent further deformity and preserve normal function. At operation the exostoses were identified in the plane of

the interosseous membrane and removed with difficulty from the parent bones. Macroscopically the specimen (Fig. 3) showed the bony processes ensheathed in fibrous tissue continuous with the radial and ulnar periosteum at their bases and with the interosseous membrane at their edges. Removal of the fibrous tissue disclosed a minute cartilage-covered " condyle " (radial exostosis) articulating with the cartilage-lined " acetabulum " (ulnar exostosis).

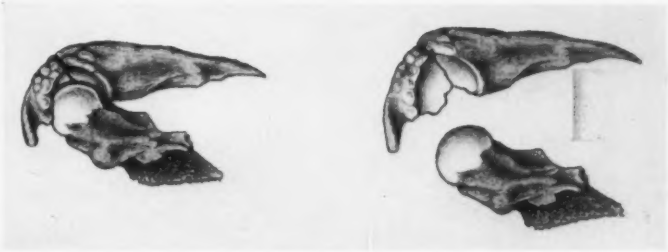

FIG. 3
Macroscopic appearances of the excised bony processes.

Progress—The radius and ulna have since grown normally (Fig. 2) and the patient rapidly resumed ordinary use of her arm. It is to be emphasised that there was at no time any loss of function. The abnormal joint lay directly in the axis of pronation-supination of the forearm, and its plane of movement was perpendicular to this axis (Fig. 1).

Histological findings—Both the " condyle " and the " acetabulum " were covered with cartilage. There was a joint cavity between the two articular cartilages, and even a fibrous tissue covering to the joint resembling a capsule; the " acetabulum " appeared to be in two

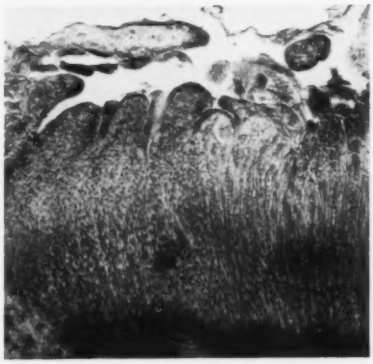

FIG. 4
Histological section of the articular cartilage of the ulnar socket showing the protrusions from its surface in the process of forming loose bodies. The nuclei of the cartilage are arranged in streamlines towards these protrusions. (H. and E., ×48.)

parts (Fig. 3). The surfaces of the articular cartilages, particularly in the " acetabulum," were not smooth; there appeared to be small finger-like protrusions of the cartilage in many places. In the joint cavity were many small loose bodies, several of which could be seen quite clearly to be in the process of formation from the processes of the articular cartilage (Fig. 4); some had become calcified, or were being calcified during extrusion from the surface of

the cartilage. The joint surfaces were not closely apposed. There was no regular arrangement of cells in the articular cartilages of the " acetabulum," the cells of which appeared to lack boundaries; consequently it appeared more like a syncitium with many nuclei arranged in streamlines which were mainly directed towards the cartilaginous protrusions from the surface. This appearance was in marked contrast to the arrangement of cartilage cells into " nests " in the articular cartilage covering the radial exostosis (Fig. 5). The cell nests were composed of cells whose nuclei were pyknotic, and the cytoplasm retracted from the cartilaginous lacunae containing them.

FIG. 5
Histological appearance of the articular cartilage covering the radial " condyle," showing the cell nests. (H. and E., ×30.)

The " capsule " of the joint was lined by a membrane resembling synovial membrane, and at one point, close to the junction of the capsule with the edge of the joint surface, there was a proliferation of subsynovial tissue to form a cellular villus-like structure projecting into the joint cavity. The nuclei in this villus were rich, and in places formed nests (Fig. 6).

The bone forming the exostoses was entirely cancellous, but at the periphery there was evidence of the formation of periosteal bone in lamellae from the well marked periosteum covering the exostoses (Fig. 7). The cancellous bone contained well developed medullary spaces containing blood vessels and a network of reticular fibres.

DISCUSSION

The question arises whether this lesion was traumatic or developmental in origin. Both the patient and her mother were emphatic that there had been no injury; but, even if this evidence is possibly unreliable, the abnormal bony processes lack the flattened, spread appearance characteristic of a traumatic pseudarthrosis and possess the shape and cartilage cover that signify a true joint, however poorly it may be formed. We believe that this condition was developmental in origin.

The similarity to congenital synostosis of the radius and ulna is also apparent, but it would be necessary to postulate an accident—which did not happen—to account for the destruction of continuity in the abnormal piece of bone; furthermore, congenital synostosis is always associated with complete limitation of pronation and supination, whereas this child had a full range of these movements. We suggest that the location of the abnormal joint

was genetically predetermined but that the action of the muscles producing movement transformed the cells from which the abnormal joint had grown into the characteristic tissues of a synovial joint. This idea may have some bearing on the developmental changes leading to the formation of a normal joint.

The mere presence of loose bodies in such an anomalous joint would in itself call for comment, but the changes associated with the formation of these loose bodies are even more remarkable. The articular cartilage of the ulnar " acetabulum " was densely populated with cell nuclei arranged in streamlines orientated towards protrusions at its joint surface which

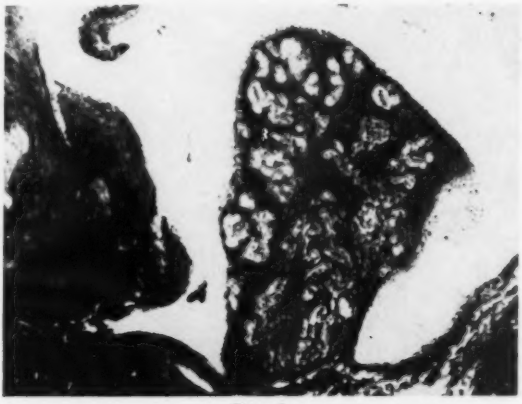

FIG. 6
Section of a synovial villus containing cell nests projecting into the joint cavity. (Masson, ×50.)

FIG. 7
The surface of the cancellous bone of the radial exostosis, showing the production of lamellae of periosteal bone (at the top of the figure). (Mallory, ×30.)

were demonstrably " loose bodies " in the process of formation, whereas the cartilage of the radial " condyle " had its cells arranged in nests and was not forming loose bodies. As in the condition of synovial chondromatosis, loose bodies were being produced by the overgrowth of living tissue rather than the shedding of dead tissue, but this case demonstrates that such a process can affect cartilage as well as synovial membrane.

Finally, there was little coaptation of the articular surfaces of the false joint, and this was associated with proliferation of articular cartilage, a feature which may have some bearing on the process of degeneration of a normal joint.

SUMMARY

An anomaly is reported in a girl aged eight years in which an outgrowth from the left radius articulated with a hollow in an eminence on the corresponding ulna. The histological findings are described and the significance of this unusual malformation is discussed.

FAMILIAL METAPHYSIAL DYSPLASIA

J. E. A. David and P. E. S. Palmer, Bulawayo, Southern Rhodesia

From the Bulawayo General Hospital

Familial metaphysial dysplasia is a rare disease. We have traced only eleven recorded cases and a description of a museum specimen: there is also the appearance of a femur in a Nubian grave of the Archaic period—but, from the available information, we are not convinced that this should be included.

We record here full details of another case, with unusual features, and details of the patient's relatives, two of whom are affected. A further family with less marked changes is also reported.

The disease was first described briefly by Pyle in 1931. The same case and that of the patient's sister were later reported in detail by Bakwin and Krida in 1937. The next report was that of Hermel, Gershon-Cohen and Jones (1953). Recently two cases were described by Komins (1954), three cases by Jackson, Albright, Drewry, Hanelin and Rubin (1954), and two cases by Feld, Switzer, Dexter and Langer (1955).

The diagnosis is made radiologically, and the disease appears to have little effect on the general health of the patient. It is not progressive. It is undoubtedly familial: the findings in our case suggest that it is transmitted through the male, but Jackson *et al.* recorded a case transmitted through the mother.

The most striking change is the extreme widening of the metaphysial ends of the long bones, with marked thinning of the cortex in the widened areas: the mid-diaphysial areas of the long bones are usually within normal limits. All the long bones and the skull may be affected.

In some of the previous cases there has been osteomalacia and a tendency towards abnormal fragility, but in our cases there was no evidence of this. The histological findings were described by Ingalls (1933), who examined a museum specimen from a man of thirty-three years who died of tuberculosis. (This same case was briefly noted by Hamann in 1910.)

CASE REPORTS

Case 1 (Family 1)—A European girl (Ann) aged eight years was admitted with acute ulcerative stomatitis. The confinement was said to have been difficult, but the child was well at birth and thrived until the age of six months, when she had an attack of what was thought to be malaria. After this it was noticed that the child had developed a squint. Apart from this she had been fairly well except for dental troubles. According to the father, her " milestones " were normal, except for walking, which was delayed.

Family history—The family relationships are complicated. Both the mother and the father had been married before and there are three children of the father by a previous marriage, all boys. The mother had a previous child, also a boy. Together they had a further three children, of whom the patient was the first, and the mother has a further child by a subsequent marriage. The father is the " common denominator " in the abnormal series, the changes occurring only in his children, one by a previous marriage. All the children's available parents are healthy. Unfortunately the family history differs if obtained from the mother or the father, and since they are no longer on speaking terms it must be regarded as unreliable. One brother and one sister have similar bone changes which will be described later, and the father states that one relative on the mother's side has webbing of the toes.

Physical examination—The child was thin, and fifty-three inches tall. Her mentality was obviously retarded and her intelligence quotient was assessed at 46 by the educational psychologist. The face was small and narrow, with a rather beaked nose and enophthalmos giving a rather parrot-like appearance (Fig. 1). There was webbing of both hands and feet. The orbits were shrunken: the action of the left superior rectus was reduced—possibly a mechanical defect rather than neurological. There was an apparent internal strabismus, simulated because of the rather broad nose: there was bilateral microcornea with small anterior chamber, and both eyes had persistent pupillary membrane with some opacity of the posterior lens capsule on both sides. We give these eye changes in detail because they

FIG. 1
Case 1—Photographs of patient.

are suggestive of arrested development at the sixth intra-uterine month, except for the posterior lens opacities which are likely to be post-natal; since there is binocular fixation these probably developed after three years. Apart from the above nothing abnormal was found on physical examination.

The following investigations were normal: blood counts, erythrocyte sedimentation rate, Wassermann reaction, blood calcium, blood sugar and serum alkaline phosphatase.

Radiographic examination—The child was originally referred for a chest radiograph and the abnormal ribs and clavicles were noted: the whole skeleton was then radiographed with the following findings. *Skull*—The shape was normal (Fig. 2). The vault appeared normal, but the base was greatly increased in thickness though not in density. The orbits were small, but normal otherwise. The mandible showed striking changes, giving the appearance of gross ballooning: most of the teeth, both temporary and permanent, were missing and the sockets could be seen in either jaw. The mastoids and sinuses were small; the antra were normal. The *spine* was normal in every respect. The *pelvis* and *scapulae* were normal. The *clavicles* showed marked ballooning at the inner ends (Fig. 3); they were symmetrical and

even, but the cortex was thinned. The general appearance suggested loss of normal density. The *ribs* were like the clavicles: they showed ballooning, especially at the sternal ends.

Long bones—The long bones were not typical of this disease as described originally. The

FIG. 2
Case 1—Skull.

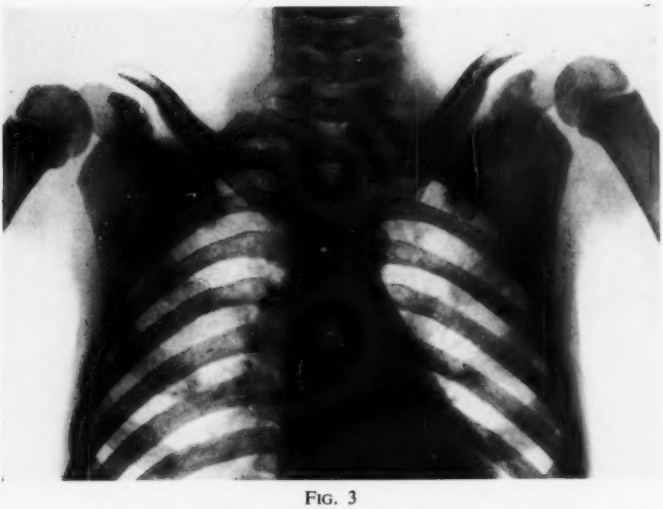

FIG. 3
Case 1—Clavicles and ribs.

mid-shafts of the radii and ulnae were widened and these bones showed generalised enlargement (Fig. 4). The shafts of the other long bones were normal (Fig. 5).

Metatarsals, metacarpals and phalanges—These showed the changes of the disease but the

FIG. 4
Case 1—Forearm bones.

FIG. 5
Case 1—Hip joints and femora.

FIG. 6
Case 1—Hands.

mid-shafts were also ballooned and not within normal limits (Fig. 6). There was syndactyly between the middle and ring fingers, and between the ring and little fingers, and the middle phalanx of both fifth fingers was missing; the middle phalanx of the ring finger of each hand was small and distorted.

The rate of growth and development appeared to be normal and there was no evidence of previous fractures or abnormal growth lines.

Cases 2 and 3 (Family 1)—One sister aged six years, born to the same parents, had slight changes in the forearms and legs but the skull and other bones were normal.

One half-brother aged fourteen, with the same father, had slight broadening of the lower ends of each radius and ulna, but the outline was less tubular than in the first case.

All other children in these families were normal.

FIG. 7

FIG. 8

FIG. 7
Case 4—Femoral shafts.

FIG. 8
Case 4—Right knee region.

Case 4 (Family 2)—A healthy European girl (Judy) aged sixteen was referred for radiography of her knees because of a rather vague history of " aching." The lower ends of both femora were widened and thinned (Figs. 7 and 8): they were flask-shaped as in Gaucher's disease, but the patient showed no evidence of any such illness. The clavicles were both broad and straight and the inner ends of the ribs were widened: the other bones were within normal limits.

Case 5 (Family 2)—One brother in a family of five showed similar changes: the children are all healthy, normally developed and intelligent. The parents refused radiographic examination.

DIFFERENTIAL DIAGNOSIS

The findings are so unusual and so characteristic that there should be little possibility of error provided the whole skeleton is radiographed. We were at first puzzled by the skull changes, which were not noted in the original cases reported. Hermel *et al.* (*loc. cit.*) reported lack of pneumatisation in the temporal bones and Pyle (*loc. cit.*) mentioned a prominent frontal region. Holt and Hodges (1953) mentioned three patients with familial metaphysial dysplasia in which there was skull thickening, and Neuhauser (1953) reported a further case. While this paper was in preparation Komins (1954) reported examples of the disease with skull changes, but there are several noteworthy differences: Komins reported a large head, eyes far apart, increased density of the skull base, and hyperostosis and dental caries; whereas

TABLE I

COMPARISON BETWEEN AUTHORS' CASES OF FAMILIAL METAPHYSIAL DYSPLASIA AND ELLIS'S CASES OF OSTEOPETROSIS

Similarities of Ellis's cases
Familial
Shape of femora and tibia (particularly in " Edward ")
Shape of radius and ulna (particularly radius)
Shape of phalanges and possibly metacarpals
Shape of skull base
Facial expressions of " Edward "
Differences in Ellis's cases
Thickening of cortex (and of base of skull and long bones)
Stippling of epiphyses
Narrowing of medulla in mid-shaft
Optic atrophy
Enlarged vault and open fontanelles; bossing of skull
Small stature

our patient had a small narrow face, enopthalmos with the eyes close together, increased thickness of the skull, but with no increase in density and no teeth. Jackson *et al.* (*loc. cit.*) recorded two examples of metaphysial dysplasia with sclerotic overgrowth of the bones of the face and base of skull resembling the changes of leontiasis ossia. He believed that the condition presented a characteristic and specific entity which he termed craniometaphysial dysplasia, and he suggested that Neuhauser's case should be placed in this group.

Similar changes and bone shapes may occur in other conditions, particularly in the femur in Gaucher's disease and in the leukaemias. In Gaucher's disease the changes are limited to the femora, and although other bones may be affected in leukaemia it is the femora that show the changes most clearly. The clubbing of the long bones may be seen in less severe types of osteogenesis imperfecta, particularly the delayed variety (Brailsford 1948), but there is a marked associated deformity and usually well developed callus remaining from previous multiple fractures; there are changes in the skull, spine and pelvis, and the mid-shafts are slender and deformed; usually the phalanges are normal. In young children, infantile

cortical hyperostosis in its healing stage might cause confusion, but the mid-shafts are widened, and usually some callus or buttressing remains. Since no case of familial metaphysial dysplasia has been recorded at the age at which infantile cortical hyperostosis occurs, the diagnosis should not present difficulty. Hypothyroidism in young children may cause " clubbing " but the clinical appearances are different and the spine is also affected. Diaphysial aclasis can produce widening of the metaphyses, but a review of the whole skeleton will make the differential diagnosis straightforward.

There is an interesting similarity in some respects between these cases and some cases of osteopetrosis (Albers-Schönberg disease), particularly in the cases described by Ellis (1934). The similarity in shape of the bones is as striking as the difference in the density. Clearly the same growth factors are disturbed, but there is a further abnormality of calcium metabolism; a comparison of the similar and dissimilar features will demonstrate this (Table I).

Ellis himself described his two cases as being atypical in some respects, particularly in that dense bone appeared in the mid-shaft and not at the growing diaphysis. This is quite the reverse of the distribution in Albers-Schönberg disease, and it is possible that Ellis's cases were examples of familial metaphysial dysplasia, particularly as Brailsford (1948) noted that the patients were radiographed again in 1938 and that the findings were not typical of osteopetrosis. In the same paragraph he drew attention to the case described by Pyle, and did not associate Ellis's cases with this condition. Possibly Ellis's cases fall midway between the two diseases: in fact, Jackson *et al.* placed Ellis's cases into their group of " cranio-metaphysial dysplasia."

The femora in our Case 1 (Ann) are similar in shape to those illustrated by Fairbank (1951) in his description of osteopetrosis, but neither of the examples is really like those originally described by Pyle. We consider that at the present time it may be premature to subdivide metaphysial dysplasia in this way when so few cases have been described.

So far all the published cases of familial metaphysial dysplasia have factors in common, mainly those of shape; and differences, mainly those of density. The study of further cases will provide a clearer picture of the condition and its numerous variants.

SUMMARY

1. Familial metaphysial dysplasia occurring in two families is recorded.
2. One case with unusual skull changes and webbing of the digits is described in detail.
3. The similarity between the shape of the bones in familial metaphysial dysplasia and other bone disease is discussed.

We are indebted to Dr P. D. G. Quirke for the ophthalmic details, Mr J. Dowley for the intelligence tests, Dr B. Tulloch for the haematological and biochemical reports and Dr J. P. Hederman, senior radiologist in Bulawayo, for his help and advice. Sir H. A. T. Fairbank has been kind enough to read and criticise this report. One of us (P. E. S. P.) acknowledges the permission of the Secretary of Health for the Central African Federation to publish these cases.

REFERENCES

BAKWIN, H., and KRIDA, A. (1937): Familial Metaphysial Dysplasia. American Journal of Diseases of Children, **53,** 1,521.

BRAILSFORD, J. F. (1948): The Radiology of Bones and Joints. Fourth edition, p. 560. London: J. & A. Churchill Ltd.

ELLIS, R. W. B. (1934): Osteopetrosis. Proceedings of the Royal Society of Medicine (Section for the Study of Disease in Children), **27,** 1,563.

FAIRBANK, Sir H. A. T. (1951): Atlas of General Affections of the Skeleton, Case 24, p. 47, Fig. 71. Edinburgh and London: E. & S. Livingstone Ltd.

FELD, H., SWITZER, R. A., DEXTER, M. W., and LANGER, E. M. (1955): Familial Metaphyseal Dysplasia. Radiology, **65,** 206.

HAMANN, C. A. (1910): An Abnormality in the Form of the Femur. Cleveland Medical Journal, **9,** 710.
HERMEL, M. B., GERSHON-COHEN, J., and JONES, D. T. (1953): Familial Metaphyseal Dysplasia. American Journal of Roentgenology, **70,** 413.
HOLT, J. F., and HODGES, F. J. (1953): The Year Book of Radiology (1953–1954 Year Book Series), p. 87. Chicago: The Year Book Publishers, Inc.
INGALLS, N. W. (1933): Bone Growth and Pathology as Seen in the Femur (and Tibia). VII. Studies on the Femur. Archives of Surgery, **26,** 787.
JACKSON, W. P. U., ALBRIGHT, F., DREWRY, G., HANELIN, J., and RUBIN, M. I. (1954): Metaphyseal Dysplasia, Epiphyseal Dysplasia, Diaphyseal Dysplasia, and Related Conditions. Archives of Internal Medicine, **94,** 871.
KOMINS, C. (1954): Familial Metaphyseal Dysplasia (Pyle's Disease). British Journal of Radiology, **N.S. 27,** 670.
NEUHAUSER, E. B. D. (1953): Growth, Differentiation, and Disease. Caldwell Lecture, 1952. American Journal of Roentgenology, **69,** 723.
PYLE, E. (1931): A Case of Unusual Bone Development. Journal of Bone and Joint Surgery, **13,** 874.

" IDIOPATHIC " SCOLIOSIS OF CONGENITAL ORIGIN

A. H-Ros Codorniu, Madrid, Spain

From the Gran Hospital de la Beneficencia General, Madrid

The etiology of idiopathic scoliosis awaits clarification, but it may be that in some cases congenital factors are responsible for the spinal deformity (Etienne 1953, Roederer 1954). The present report of " idiopathic " scoliosis in identical twins gives further support to these views.

CASE REPORT

The patients were identical twins thirteen years old, in whom until three years before no spinal abnormality had been detected by the mother, the family doctor or the girls themselves. At the age of ten a progressive scoliosis appeared, starting in both girls at exactly the same time and presenting from the beginning similar characteristics and rate of deterioration (Fig. 1).

Fig. 1
The twins, C on the left and P on the right, showing the similarity of the spinal curves.

The radiographs show a striking identity between the two spines (Fig. 2), and even the amount of correction obtainable by traction appears alike in both cases. No hemivertebra, spina bifida or any other congenital anomaly is detectable in these radiographs.

The genetic study of these two girls is summarised in Table I. The twins were born with a single placenta, and it seems beyond doubt that they were uniovular.

DISCUSSION

Genotypic or phenotypic disorders of the spine such as hemivertebra, fusions and rachischisis appear with the secondary scolioses, but this is never the case with idiopathic scoliosis, in which for years no deformity or anomaly of any kind has been detected in the

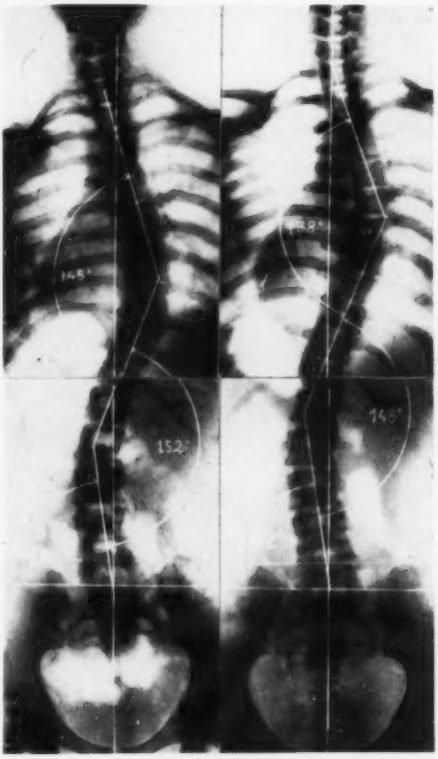

FIG. 2
Radiographs of the spines, C on the left and P on the right.

spine. This suggests that at the beginning of growth the fact or factors responsible for the deformity are either not present or not acting, or that at least their effect, if any, is not felt. Another interesting finding in the radiographs of young people suffering from scoliosis is that the deformed vertebral bodies show greater inclination in their cephalic than in their caudal segment. These late changes may perhaps be due to some particular anomaly of the blood flow of the vertebral body rather than to any embryonic stigma, a possibility which would explain the observation by Kleinberg (1926) of larger vascular foramina in the vertebral bodies in scoliosis on the side of the convexity than on the side of the concavity. Unfortunately, at present we do not know whether this deformity precedes or follows the vertebral inclination.

That some types of "idiopathic" scoliosis may be due to factors genetically determined is now beginning to be accepted by a number of authors. Kleinberg (*loc. cit.*) considered that about 25 per cent of all cases may have such an origin. We have treated a family in which the father and three of their six children suffered from this deformity.

TABLE I

CHARACTERISTICS OF THE TWINS

	Twin C	Twin P
Blood group	AB	AB
Height	147 centimetres	148·5 centimetres
Weight	34 kilograms	40 kilograms
Colour: eyes	12–13	12–13
hair	8	8
skin	White	White
Type of hair	Straight	Straight
Hair distribution	Alike	
Tongue sulcus	Alike	
Shape of ears	Alike	

A detailed study of the blood supply of the spine in all its segments and at different stages of evolution until its final fusion at the end of growth seems urgently needed to elucidate many of these still obscure points.

I wish to thank Dr Jimena de la Vega for the genetic studies of these cases.

REFERENCES

ETIENNE, E. (1953): La scoliose dite "essentielle." Revue de Chirurgie Orthopédique, **39**, 602.
KLEINBERG, S. (1926): Scoliosis. New York: Paul B. Hoeber.
ROEDERER, C. (1954): Le traitement orthopédique de la scoliose. Sixième Congrès International de Chirurgie Orthopédique Berne, 30 août–3 septembre 1954. Procès-verbaux, &c., p. 242. Bruxelles: Imprimerie Lielens.

IDIOPATHIC SCOLIOSIS IN IDENTICAL TWINS

RAFAEL ESTEVE, BARCELONA, SPAIN

From Camitas Blancas (Instituto Policlinico) and Hospital de San Juan de Dios, Barcelona
Formerly at the Nuffield Orthopaedic Centre, Oxford

The etiology and pathogenesis of " idiopathic " scoliosis are still to be elucidated. It is generally agreed that scoliosis is a condition related to growth, for it does not increase appreciably after the end of growth. Thus imbalance of the spinal muscles appearing after growth has ceased produces little or no lateral curvature. A primary disturbance of the vertebral growth centres has not yet been proven, but it is thought that, once a lateral curve has developed, the imbalance in the distribution of stress on the epiphysial plates increases the deformity following the laws of epiphysial growth (Hueter 1862, Volkmann 1872) and the recent studies of Trueta (1957). Steindler (1951) stated that " the fact that the deformity develops under gravitational influence implies that it is selective, and this again means that certain predisposing factors must be at work."

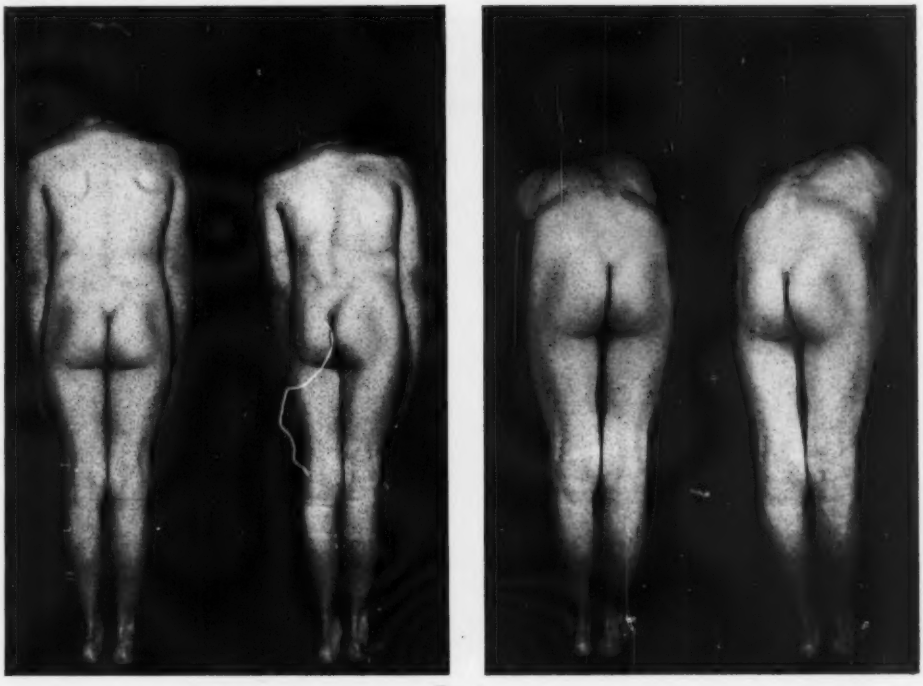

FIG. 1
Photographs of the twin girls showing the similarity of the spinal curves. The scoliosis is more marked in S (right).

The nature of that predisposing or primary factor is not known. A list of the possible local factors should include anatomical alterations in the growth nucleus or the epiphysial plates, in the pattern of vascularisation of the vertebrae, alterations in the spinal muscles or their innervation, and also metabolic or endocrine disturbances. Any of those disturbances

might have a congenital origin and therefore be predetermined or present at birth, though only developing later in life like many other congenital abnormalities.

In this respect it is of interest to report this example of idiopathic scoliosis occurring in identical twins at the same time and with similar characteristics.

CASE REPORT

The twin girls are from a family of eight children. The parents are living and healthy. There is no consanguinity, and deformities, congenital or otherwise, are unknown in the family. Nothing abnormal is known to have occurred during the mother's pregnancy or at parturition.

FIG. 2
Radiograph of the spine of twin P.

FIG. 3
Radiograph of the spine of twin S.

The girls, to whom I shall refer as P and S, were seen by me at eleven years of age. At the age of six a curvature of the spine, with prominence of the right scapula, was noticed in both girls, being slightly less marked in P. Spinal supports have been worn since the age of seven.

On examination the twins were alike. The colour of the eyes (dark brown), of the hair (dark brown) and of the skin (light), the shape of the ears and the distribution of hair were identical. They were alike in build, but S was an inch and a half shorter than P because of the greater spinal curvature. The length of the upper and lower limbs was equal on both sides and in both girls. There was thoracic scoliosis convex to the right and lumbar scoliosis

convex to the left, more marked in S (Fig. 1). Neurological examination was negative. There was no café-au-lait marking.

Radiographic examination—In P there was a thoracic scoliosis of 46 degrees convex to the right, with the apex at T.7 and moderate rotation; there was moderate lumbar scoliosis to the left (Fig. 2). The only congenital anomaly seen in the spine was spina bifida of the first sacral segment. In S there was a thoracic scoliosis of 58 degrees convex to the right with the apex at T.7; there was lumbar scoliosis to the left; and there was marked rotation in both curves (Fig. 3). No congenital anomaly was seen in the spine except spina bifida in the fifth lumbar and first sacral segments.

DISCUSSION

This is an example of scoliosis in identical (uniovular) twins. The degree of scoliosis is different, being more severe in one of the patients, but the other characters of the curves are strikingly alike, both having the same pattern and the apex at the same level—T.7.

In the absence of visible congenital deformities at the level of the curves these two patients should be included in the group of so-called idiopathic scoliosis of the thoracic type (Ponseti and Friedman 1950) with a fairly early onset (juvenile variety of James (1954)). This indicates the presence of some other congenital factor influencing the production of scoliosis at a later stage in life, perhaps through the modification of the growth pattern in a limited spinal segment.

REFERENCES

HUETER, C. (1862): Anatomische Studien an den Extremitätengelenken Neugeborener und Erwachsener. Virchows Archiv für pathologische Anatomie und Physiologie, **25,** 572.

JAMES, J. I. P. (1954): Idiopathic Scoliosis. Journal of Bone and Joint Surgery, **36–B,** 36.

PONSETI, I. V., and FRIEDMAN, B. (1950): Prognosis in Idiopathic Scoliosis. Journal of Bone and Joint Surgery, **32–A,** 381.

STEINDLER, A. (1951): Post-graduate Lectures on Orthopedic Diagnosis and Indications, Vol. 2, p. 101. Springfield, Illinois: Charles C. Thomas.

TRUETA, J. (1957): The Normal Vascular Anatomy of the Human Femoral Head during Growth. Journal of Bone and Joint Surgery, **39–B,** 358.

VOLKMANN, R. (1872): Verletzungen und Krankheit der Bewegungsorgane. *In* Pitha and Billroth Handbuch der allgemeinen und speciellen Chirurgie. Band 2, Abtheilung 2. Stuttgart: F. Enke.

CYST OF AN INTRA-ARTICULAR DISC OF THE ACROMIO-CLAVICULAR JOINT

P. BERBRAYER, WINNIPEG, CANADA

From the Department of Orthopaedics, Deer Lodge Hospital, Winnipeg

Intra-articular discs or fibrocartilages are found in the knee, in the wrist, in the acromio-clavicular joint, in the sterno-clavicular joint and in the temporo-mandibular joint. Tears or laxity of the discs have been described at all these sites. Cysts of the cartilages of the knee are well recognised, especially in the lateral cartilage; there may or may not be a history of previous injury associated with cyst formation. Except in the knee, cyst formation associated with an intra-articular disc must be quite rare, for a review of the literature does not reveal any record of such a condition.

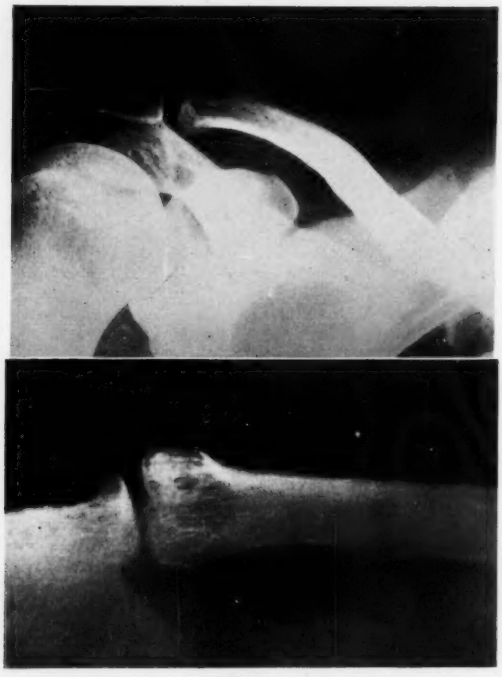

FIG. 1
Radiographs of right acromio-clavicular joint before operation.

CASE REPORT

A man aged twenty-three years had received an injury to the right shoulder when he slipped in his shower bath. A diagnosis of subluxation of the right acromio-clavicular joint had been made, and the joint was supported in plaster. A year and a half later he noticed discomfort in the right shoulder. Shoulder movements were full. A small bony prominence

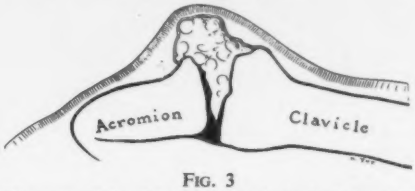

FIG. 3
Diagram of the right acromio-clavicular joint with the cyst of the intra-articular disc.

FIG. 2
Diagrammatic appearance of the shoulder as seen at operation.

FIG. 4
Photograph of the cystic intra-articular disc removed at operation.

FIG. 5
Figure 5—Histological section of the intra-articular disc. (×35.)

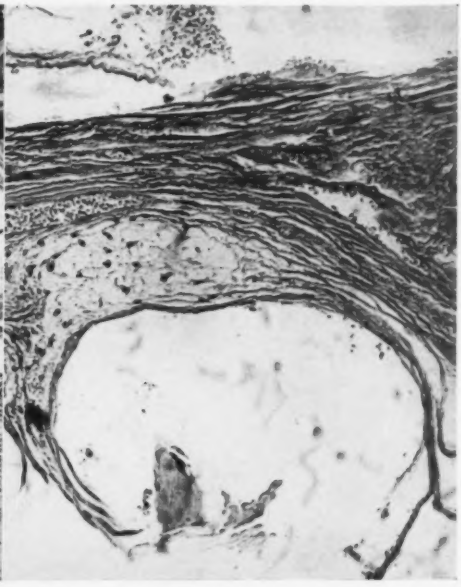

FIG. 6
Figure 6—Higher power photomicrograph of the disc. (×100.)

was noted at the outer end of the right clavicle. Radiographs of the acromio-clavicular joint, taken with and without weights, did not show any abnormality.

He attended again two years later, complaining of a slight ache over the top of the right shoulder after use. Examination showed a hard lump, about one inch by a half inch, over the region of the right acromio-clavicular joint. There was no local tenderness and no adventitious bursa in the region of the lump. There was a full range of painless movements of the shoulder. Radiographs showed slight dorsal exostoses on the articular margins of the acromio-clavicular joint (Fig. 1), but these were not large enough to account for the size of the hard lump observed clinically.

Operation—The right acromio-clavicular joint was explored. A hard, firm lump overlay its upper aspect (Figs. 2 and 3). The mass was bluish and nodular, and seemed to arise from the upper aspect of the acromio-clavicular joint and to be connected with the intra-articular disc. It was excised together with the disc (Fig. 4). The small exostoses of the superior articular margins of the joint were chiselled off.

Histological examination (Dr N. Crowson)—Section of the cystic cartilaginous structure showed fibrocartilage with some areas of degeneration and cyst formation (Figs. 5 and 6). The cysts were lined by flattened synovial-type cells. There was a mucinous type of change in the connective tissue about the cysts.

I wish to thank Dr I. Sutton, Medical Superintendent, for permission to publish this case, and Dr W. B. MacKinnon, Head of the Orthopaedic Department, and the other members of the orthopaedic staff, for their assistance in presenting the case.

THE EFFECT OF INTERMITTENT CORTISONE ADMINISTRATION IN THE RABBIT

Induction of Cycles of Resorption and Deposition of Bone

ELSDON STOREY, MELBOURNE, AUSTRALIA

From the Department of Pathology, University of Melbourne

It was found during studies on the movement of teeth with orthodontic appliances (Smith and Storey 1952, Storey 1953) that variations in the rate of tooth movement, related to the menstrual cycle, occurred in girls (Storey 1954). It was postulated that this cyclic variation in rate of tooth movement was, in part at least, related to cyclic changes in hormonal secretion affecting bone and connective tissues. For this reason study was made of the effect of producing cycles of bone changes by the use of intermittent administration of cortisone.

Previous work had shown that administration of cortisone to the rabbit is followed by rapid resorption of bone (Storey 1957*a*); and, after its withdrawal, an equally rapid deposition of bone occurs, not only in the resorbed areas but on periosteal surfaces and within sutures (Storey 1957*b*). The new bone is separated from the old by a darkly staining haematoxylin " reversal " line which, on the periosteal surface of the skull, resembles a layer of bone formed by appositional growth. It is known that periosteal activity during normal (Weinmann and Sicher 1955) or neoplastic growth (Sissons 1949) is also intermittent, as shown by successive rings of bone—particularly on the long bones and the vault of the skull. However, whether these superimposed layers of bone are local in origin or are local manifestations of general hormonal control is not known.

Jaffe, Bodansky and Blair (1932), by injecting rabbits with various doses of parathormone in an attempt to prolong its effect without killing the animals, produced cyclic changes in bone structure. They demonstrated a small series of " reversal " lines in the bone surrounding vessels in the skull; but they made no mention of any periosteal activity.

If normal growth processes in bone are influenced by the cyclic changes of endocrine activity it should be possible, in any attempt to study bone pathology, to induce cycles of bone change that are abnormal in intensity and duration as well as those that are normal. For instance Jaffe (1933) ascribed the change in osteitis deformans to an increased cycle of resorption and deposition of bone which gives rise to the typical mosaic pattern shown in this condition. Selye (1932) also produced bone changes of a different character by administering small doses of parathormone every two days: paradoxically, instead of bone resorption, an increased bone deposition took place.

This present paper is concerned with a preliminary study of cycles of resorption and deposition of bone in the rabbit after intermittent cortisone administration. The results have been used as a basis for a large-scale, long-range experiment now in progress.

EXPERIMENTAL METHOD

Cortisone acetate was administered to rabbits—eight young (three to five months) and one old (three years)—in doses of twenty-five milligrams a day for various periods; it was then withdrawn for a further period. A period of administration followed by withdrawal of the hormone will be referred to as one cycle. Series of one, two, three and four cycles of cortisone administration were produced in a number of rabbits. The time intervals of the various cycles are shown in Table I.

Coronal sections of the skull were cut an eighth of an inch behind the coronal suture; specimens were also obtained from the femur and premaxillae. They were decalcified in

TABLE I

TIME INTERVALS OF CORTISONE ADMINISTRATION

(+ refers to administration of cortisone)
(− refers to withdrawal of cortisone)

Cycle	Cortisone administration (*in days*)	Number of animals
1	+7 −2	1
	+7 −7	2
2	+7 −7, +7 −7	1
3	+7 −14, +7 −7, +7 −7	1
4	+7 −7, +7 −7, +7 −14, +5 −3	2

5 per cent nitric acid; and histological sections were cut at 10μ and stained with Ehrlich's acid haematoxylin and eosin.

RESULTS

Normal bone—The bones of normal male and female rabbits of ages corresponding to those in the experiment were examined. In the skull " reversal " or " arrest " lines are found both on the outer and inner plates of the cranial bones, where they run a relatively straight course parallel to the periosteal surface. They vary considerably in numbers in the young animals whereas in the older animal there are more. These lines are not present in the bone adjoining the sagittal or premaxillary sutures; a few are found on the outside of the middle of the shaft of the femur.

INTERMITTENT ADMINISTRATION OF CORTISONE

One cycle (+7 −2 and +7 −7)—The administration of cortisone for seven days followed by two days' withdrawal is associated with resorption of bone of the skull, but the usually observed increases of vascularity and appearance of multinucleated osteoclasts are absent. After seven days' withdrawal of cortisone a general change from resorptive activity to laying down of new bone is most obvious (Storey 1957*b*).

A new layer of bone has formed on the outer and inner periosteal surfaces of the skull (Fig. 1). This new layer is not continuous along the entire surface of the bone.

Within the sagittal suture there is a thickening of the blue-staining haematoxylin layer at the junction of the sutural connective tissue and bone; in places this widens to form a new thin layer of bone.

New bone also forms on the periosteal surface and within the trabeculae of the premaxilla. There is little change in the growing epiphysial cartilage of the femur; normal growth has not yet been resumed. There are numerous areas within the bone where new bone formation is occurring—a darkly staining " reversal " line separates the old and new bone.

Two cycles (+7 −7; +7 −7)—Two new layers of bone have formed on the outer and inner periosteal surfaces of the skull (Fig. 2), but in places a single layer has formed on the endosteal surface. These layers of bone are separated by two lines which sometimes converge to form one or even disappear; within the sagittal suture two lines are present. Similarly two layers of bone are seen on the periosteal surface of the premaxilla and femur. Within the bone of the skull it is possible to see where two waves of resorption and deposition have formed two " reversal " lines around some small vascular spaces.

FIG. 1

The effect of one cycle of cortisone administration on the outer plate of bone of the cranial vault in the young rabbit. A new layer of bone is formed on the periosteal surface and around vascular spaces in the bone. This layer of bone is separated from the old bone by a darkly staining haematoxylin line. (×400.)

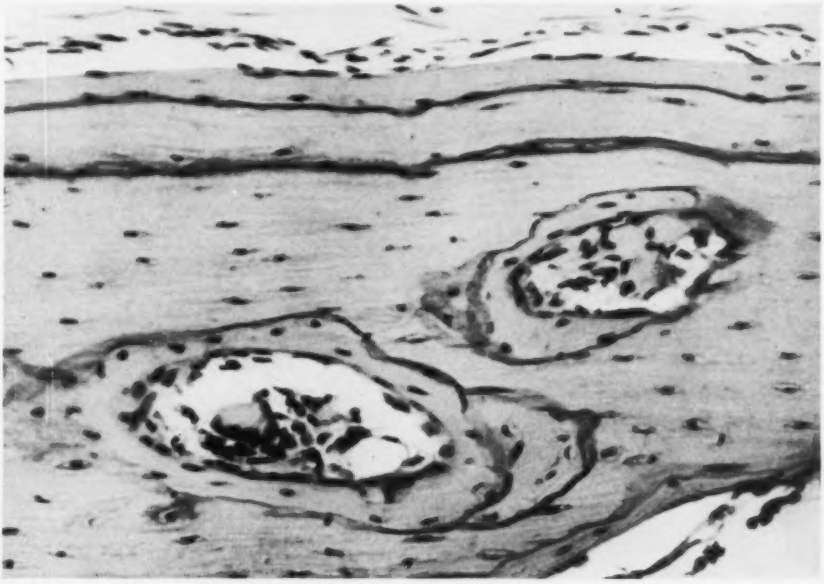

FIG. 2

The effect of two cycles of cortisone administration on the outer plate of bone of the cranial vault in the young rabbit. Two new layers of bone are formed on the periosteal surface separated by two reversal lines. The first line appears double in places. (×400.)

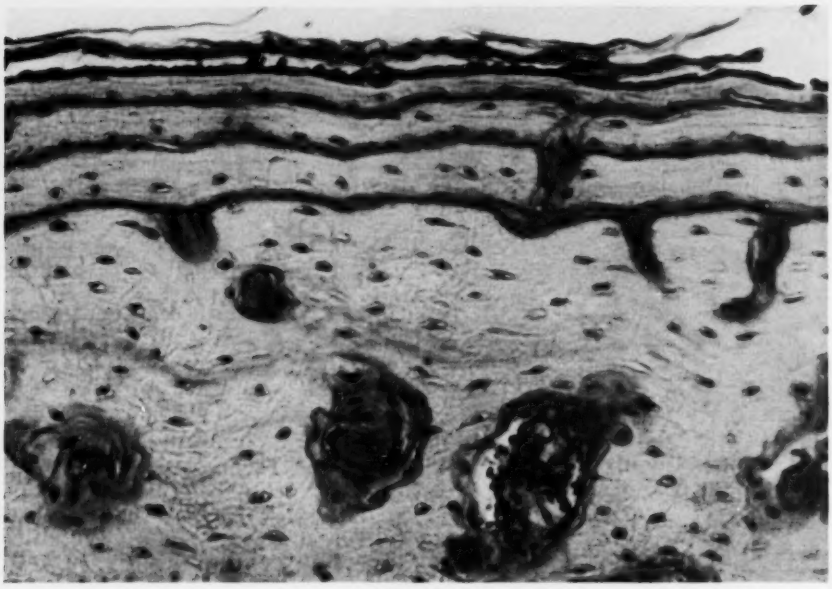

FIG. 3

The effect of three cycles of cortisone administration on the outer plate of bone of the cranial vault in the young rabbit. Three new layers of bone are formed on the periosteal surface. The first layer formed during fourteen days' withdrawal of cortisone is approximately twice as thick as the outer two. (×400.)

FIG. 4

The effect of four cycles of cortisone administration on the outer plate of bone of the cranial vault in the young rabbit. Four new layers of bone formed on the periosteal surface. The third layer formed during fourteen days' withdrawal of cortisone is twice as thick as the first two; the outer layer is very much narrower, showing the small amount of bone formed during three days' withdrawal of cortisone. The vascular spaces in the bone show areas of resorption in this particular section. (×400.)

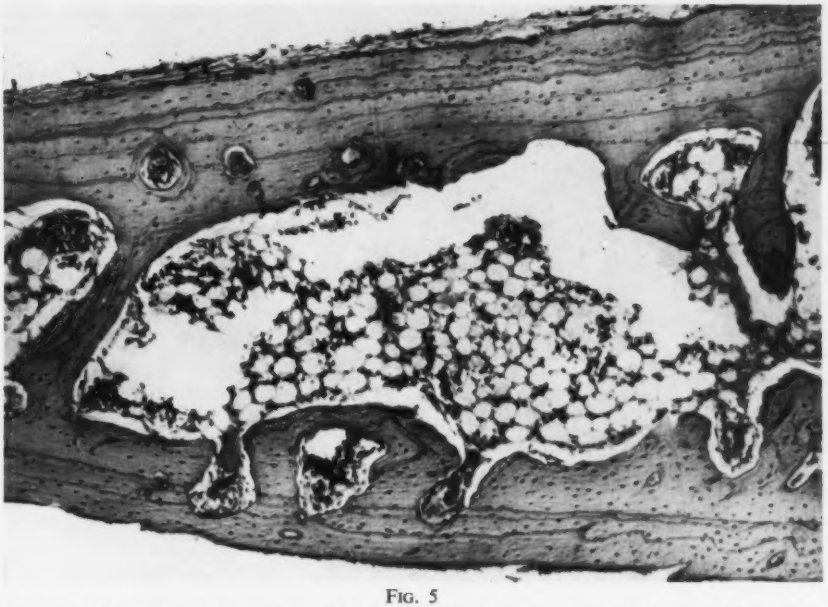

FIG. 5

Coronal section of cranial vault showing the formation of four new layers of bone on both external and internal surfaces after four cycles of cortisone administration in the young rabbit. The fourth layer of new bone has been formed only partly in this section but is shown more clearly in Figures 4 and 7. (×80.)

FIG. 6

Area adjacent to the section shown in Figure 5. There are four cycles of cortisone administration. Associated with the new layers of appositional bone growth there are changes in the internal architecture of the trabeculae, which are irregularly arranged and contain numerous "reversal" lines. The fourth layer of new bone is clearly apparent only at the upper right-hand corner. (×240.)

FIG. 7

The effect of four cycles of cortisone administration on the anterior aspect of the cranial vault in the young rabbit. The width of new bone is greater than that formed towards the posterior aspect of the skull in the same animal, although the same sequence of layers is present (compare with Fig. 4). Note the accelerated reconstruction of the underlying bone and the appearance of irregular reversal lines. (×240.)

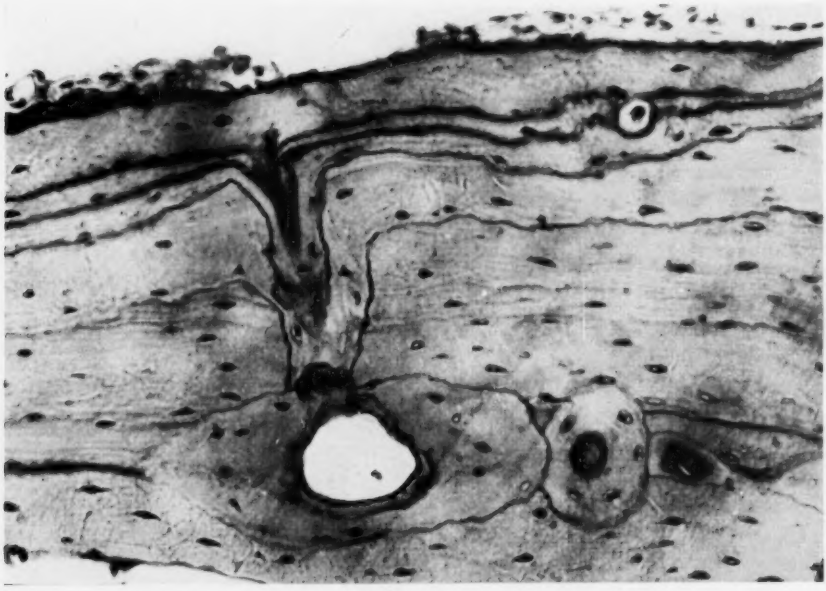

FIG. 8

The effect of four cycles of cortisone administration in the old animal. The width of new bone formed is less than in the young animal although the same sequence of layers is seen. The fourth layer is only just detectable towards the upper left part of the section (compare with Figs. 4 and 7). (×400.)

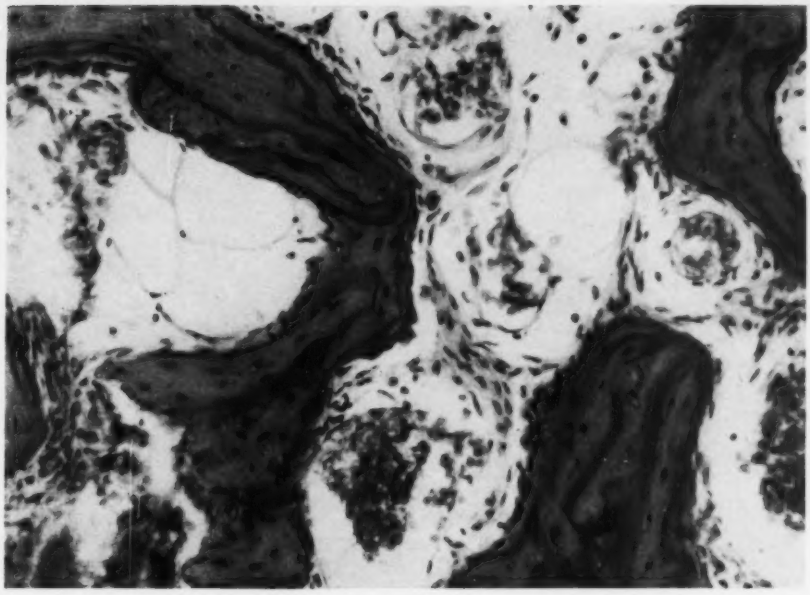

FIG. 9

The effect of four cycles of cortisone administration on the structure of the bone trabeculae. Repeated waves of resorption and deposition have left a series of irregular reversal lines in the bone structure. The marrow spaces are filled with greatly dilated vessels, a few fat cells and young connective tissue. (×240.)

FIG. 10

Higher magnification of the section shown in Figure 9. A bone trabecula shows the increased number of new reversal lines and new bone formation. (×1,000.)

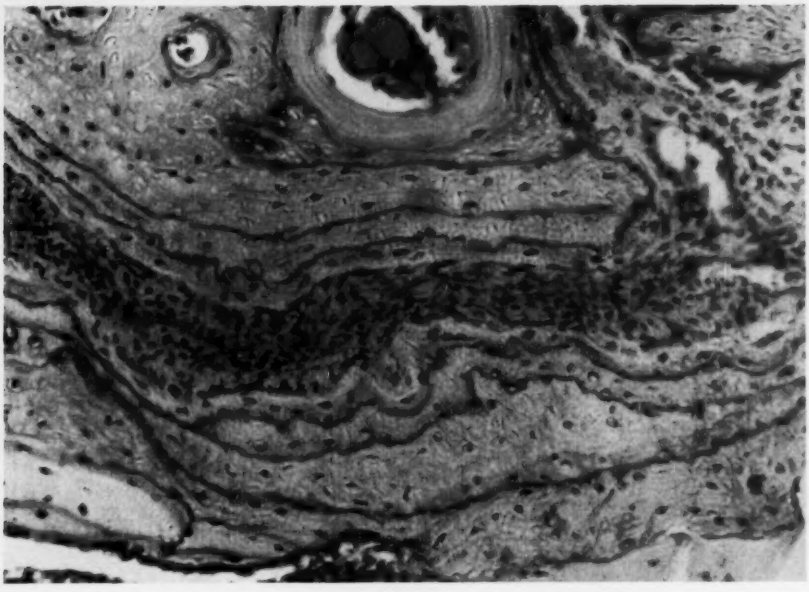

FIG. 11

The effect of three cycles of cortisone administration on the bone adjoining the sagittal suture. The wide layers of bone corresponding to fourteen days' withdrawal of cortisone are farthest from the sutural connective tissue. (×240.)

FIG. 12

The effect of four cycles of cortisone administration on the structure of Haversian systems: repeated " reversal " lines show where interruptions of bone formation occurred. In the lower left corner of the section three " reversal " lines converge to form a single thick line and then continue as two lines. (×1,000.)

FIG. 13

The effect of four cycles of cortisone administration on the epiphysis of the lower end of the femur. The metaphysial cartilage is replaced by a thin layer of dense bone. (×80.)

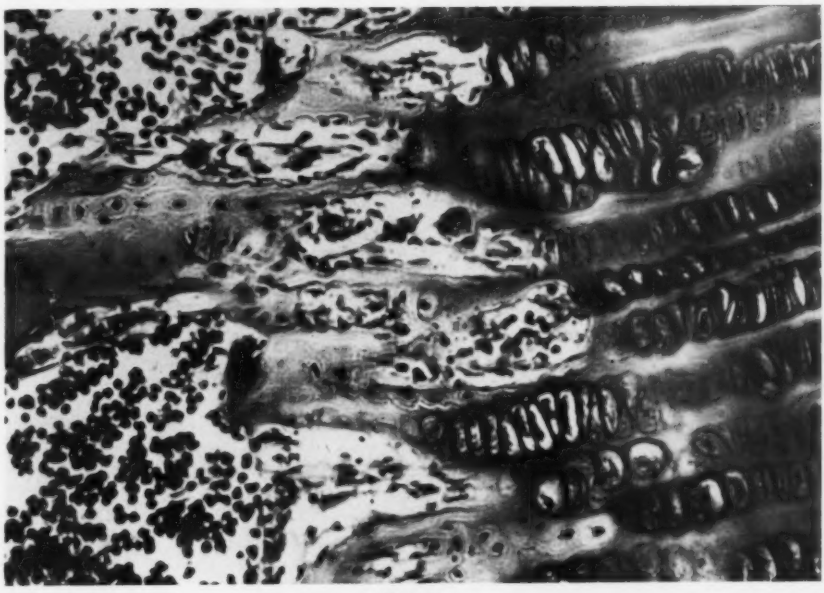

FIG. 14

Higher magnification of Figure 13. Formation of stunted bone trabeculae at the end of the irregularly arranged epiphysial cartilage cells. (×240.)

Three cycles (+7 −14; +7 −7; +7 −7)—Three new layers of bone have formed on the outside and inside surfaces of the two cortical plates of the skull (Fig. 3). The new bone corresponding to the first cycle is two to three times as wide as the second and third layers. These layers of new bone are present also in the sagittal suture (Fig. 11), on the outer periosteal surface of the premaxilla, and towards the middle of the shaft of the femur. They show the same pattern; that is to say, the first layer formed is two to three times as wide as each of the second and third layers of bone.

The internal architecture of the bone is changing—around some vascular spaces there is a number of lines showing where repeated resorption and deposition have taken place.

Four cycles (+7 −7; +7 −7; +7 −14; +5 −3)—A young as well as an old animal were included in this experiment.

In the young animal four layers of new bone have formed on the outside and inside periosteal surfaces (Figs. 4 to 6) and in places within the sagittal and coronal sutures. The width of the new layers of bone varies according to the time for which cortisone had been withdrawn; the thickest layer is formed during fourteen days, the thinnest during three days. This last layer is not present in all sections; it is absent near the coronal suture and becomes prominent only towards the front of the cranial vault (Fig. 7). The thickness of the haematoxylin " reversal " line does not vary in any of these cycles, although the lines are much thicker and more darkly staining than those formed in the bone during the previous normal growth of the animal.

Similar new layers of bone have formed on the periosteal surfaces of the premaxilla and towards the centre of the shaft of the femur. These new layers of bone show the same distribution of width between the " reversal " lines.

The internal architecture of the bone, particularly in the skull, is beginning to change appreciably (Figs. 6 and 9). Some vascular spaces widen, showing increased vascularity and the presence of small osteoclasts at resorbing bone edges. Others show Haversian systems in which three or four concentric " reversal " lines are present around the central blood vessel (Fig. 12). Some trabeculae show an increased number of irregularly arranged " reversal " lines which correspond to the cycles of resorption and deposition (Fig. 10). The trabeculae are becoming irregularly arranged, and some of the marrow spaces are filled with extremely vascular connective tissue.

Significant changes have now taken place in the growing epiphysis of the femur (Figs. 13 and 14). There is a disappearance of long columns of metaphysial calcified cartilage; instead, a layer of new bone, which partly seals the epiphysial cartilage from the marrow space, is forming. The epiphysial cartilage is irregular in outline; cartilage cells vary greatly in development at the same level and some are enclosed in the newly formed bone.

The same pattern of new appositional bone growth seen in the young animal is shown in the old animal, but the changes are much less obvious (Fig. 8). Appositional bone growth has occurred in some places, but not over the whole cranial vault. The lines separating these layers of new bone either converge at points to form one or two lines within the bone, or disappear completely. Within the suture much more extensive (and possibly more rapid) changes have taken place. There are no lines indicating new layers of bone; instead there are large areas of haematoxylin-staining granules. Around small vessels some changes are found, but not to the extent shown in the young animal.

DISCUSSION

The intermittent administration of cortisone in the rabbit is associated with cycles of resorption and deposition of new bone in both growing and mature rabbits. In the young animal attention is directed to new growth of bone by darkly staining haematoxylin lines partitioning it into layers. These lines are more obvious and less regular than those formed

previously in the bone. They are probably best designated as " reversal lines," because they form at the place where (and the time when) resorption is followed by deposition of bone. The width of the new layers of bone is of the same order, for the same time interval, in different animals of the same age, but in the old rabbit less bone forms. When different areas in the same animals are compared the amount of new bone varies—on the frontal aspect the line of bone is wider than on the occipital aspect of the skull, and relatively little forms in the middle of the shaft of the femur.

It is noteworthy that new layers of bone form not only on the outer, but also on the inner, aspect of the skull. The appearance of lines within the sagittal suture shows that separation of these bones must occur during growth of the skull. Indeed, the placing of lines within bone substance in this way could be used as an accurate indicator of the extent and direction of growth of skull bones.

Although the general emphasis is on active new bone formation after the withdrawal of cortisone, the influence of local factors modifies the response so that in some areas resorption of bone continues. Similarly, whilst appositional bone growth can be induced by these cycles of hormonal changes, new bone growth does not take place to the same extent in all bones. In some areas no bone forms, and in others reversal lines converge and run into one another to form single wide lines.

The internal architecture of the bone slowly changes with these repeated cycles of resorption and deposition of bone. This is particularly obvious round vessels within the bone, where waves of resorption followed by deposition leave the irregular deeply staining haemotoxylin lines in the bone substance. Bone trabeculae also show this—numerous " reversal " lines are present and there are even irregularities in the arrangement of trabeculae.

The appositional growth associated with the intermittent administration of cortisone may be due to periodic interruptions in the normal continuous growth of bones. It is known that cortisone inhibits the growth of bone in the rabbit (Sissons and Hadfield 1955) and since it is also known to influence the pituitary gland (Halmi and Barker 1952) the effect may be by way of change in the growth hormone. This may be true for young animals but, in the case of the older non-growing animal, this would not be so likely. Here it is probable that growth, after the withdrawal of cortisone, is in part due to a " rebound " phenomenon activating the endocrine systems concerned with growth. As the administration of cortisone is associated with atrophy of the adrenal cortex (Ingle and Mason 1938) its rapid withdrawal probably leads to a relative deficiency of cortical secretion. This temporarily induced " medical adrenalectomy " may be associated with the observed overgrowth of bone, as adrenalectomy in the well-fed rat is followed by an increased epiphysial growth (Wyman and Tum-Suden 1945) and accelerated regrowth of hair (Zeckwer 1953). Both of these observations are ascribed to increased secretion of growth hormone.

There are changes at the epiphyses of the growing bones following these cycles of resorption and deposition. A new layer of dense bone is forming which already partly seals off the epiphysial cartilage from the marrow space. This change differs from that seen after the administration of A.C.T.H. and cortisone in the rabbit (Storey 1957*b*).

It is noteworthy that at the time that cartilaginous growth is slowing down, thickening of the skull and long bones is continuing by an appositional process. These two changes, to some extent, are parallel to those normally seen during the growth and maturation of the limbs of the skeleton, where growth in length occurs first and is followed by thickening of bones (Silberberg and Silberberg 1941). It is perhaps relevant to ask whether the maturation of bone is due to changes not only in quantity and type of endocrine secretion but also to an alteration from a more or less continuous to an intermittent or cyclic production of hormones. Such alterations in rates of endocrine activity are already known to occur during the sexual development of girls (Nathanson, Towne and Aub 1941).

It is clear that a wide variation of bone changes can be produced by the use of cortisone

in the rabbit. The continuous administration of cortisone leads to a general osteoporosis of the skeleton, whereas intermittent administration is associated with appositional growth of the skull and changes in the pattern of the bone structure; these are characterised by an intense increase of vascularity and the appearance of darkly staining wide irregular " reversal " lines in the bone substance. It is possible that by altering the duration of these cycles variations of this change may be produced, ranging from rarefying to sclerosing conditions of bone. Also, since stress areas are particularly susceptible to continuous cortisone administration (Storey 1957*a*), the localisation of these changes to areas of bone under abnormal stress is to be expected. Experiments dealing with the long-range effects of such cycles of resorption and deposition of bone are now in progress and will be reported later.

These bone changes, associated with increase in the amount of cortical adrenal hormone, are specific and resemble changes seen in naturally occurring diseases. These diseases may well be due to such influences. It is not intended at this stage to convey, however, that they are necessarily due to changes in the adrenal structure or secretion. Indeed, they may be due to the aberrant action of other ductless glands; they may well be due to various combinations of ductless gland activity.

Such hormonal disturbances may be " primary," but more likely they are themselves caused by (or are associated with) other disturbances of the vascular or nervous system or by other factors. The important point is that basic observations of the type given here form an essential groundwork for the elucidation of the more complex changes of naturally occurring diseases, without necessarily giving any immediate clue to their doubtless motley and protean pathogenesis.

SUMMARY

The intermittent administration of cortisone in both the young and the mature rabbit is associated with appositional bone growth on the periosteal surfaces of the cranium, premaxilla and middle of the shaft of the femur; each new layer of bone is separated from the next by a darkly haematoxylin-staining " reversal " line. The internal architecture of the bone also changes in consequence of the repeated waves of resorption and deposition of bone round vascular spaces.

Cartilaginous growth at the epiphysis in the young rabbit is also affected. The long columns of metaphysial cartilage are replaced by a layer of new bone which partly seals the epiphysial cartilage from the marrow spaces.

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REFERENCES

HALMI, N. S., and BARKER, S. B. (1952): Histo-physiological Effects of Cortisone on Rat Pituitary and Thyroid. Endocrinology, **51,** 127.

INGLE, D. J., and MASON, H. L. (1938): Subcutaneous Administration of Cortin Compounds in Solid Form to the Rat. Proceedings of the Society for Experimental Biology and Medicine, **39,** 154.

JAFFE, H. L. (1933): Paget's Disease of Bone. Archives of Pathology, **15,** 83.

JAFFE, H. L., BODANSKY, A., and BLAIR, J. E. (1932): The Influence of Age and of Duration of Treatment on the Production and Repair of Bone Lesions in Experimental Hyperparathyroidism. Journal of Experimental Medicine, **55,** 139.

NATHANSON, I. T., TOWNE, L. E., and AUB, J. C. (1941): Normal Excretion of Sex Hormones in Childhood. Endocrinology, **28,** 851.

SELYE, H. (1932): On the Stimulation of New Bone Formation with Parathyroid Extract and Irradiated Ergosterol. Endocrinology, **16,** 547.

SILBERBERG, M., and SILBERBERG, R. (1941): Effects of Hormones on the Skeleton of Mice, Guinea Pigs and Rats. Endocrinology, **29,** 475.

SISSONS, H. A. (1949): Intermittent Periosteal Activity. Nature, **163,** 1,001.

SISSONS, H. A., and HADFIELD, G. J. (1955): The Influence of Cortisone on the Structure and Growth of Bone. Journal of Anatomy, **89**, 69.

SMITH, R., and STOREY, E. (1952): The Importance of Force in Orthodontics. The Design of Cuspid Retraction Springs. Australian Journal of Dentistry, **56**, 291.

STOREY, E. (1953): Bone Changes Associated with Tooth Movement. A Radiographic Study. Australian Journal of Dentistry, **57**, 57.

STOREY, E. (1954): Bone Changes Associated with Tooth Movement. The Influence of the Menstrual Cycle on the Rate of Tooth Movement. Australian Journal of Dentistry, **58**, 80.

STOREY, E. (1957*a*): The Influence of Cortisone and A.C.T.H. on Bone Subjected to Mechanical Stress (Tooth Movement). Submitted for publication.

STOREY, E. (1957*b*): The Effect of Continuous Administration of Cortisone and Its Withdrawal on Bone. Australian and New Zealand Journal of Surgery, **27**, 19.

WEINMANN, J. P., and SICHER, H. (1955): Bone and Bones. Second edition. St Louis: The C. V. Mosby Company.

WYMAN, L. C., and TUM-SUDEN, C. (1945): The Effect of Adrenalectomy on the Epiphyseal Cartilage of the Rat. Endocrinology, **36**, 340.

ZECKWER, I. T. (1953): The Accelerating Effect of Adrenalectomy on Regrowth of Hair in the Thyroidectomised Rat. Endocrinology, **52**, 39.

PROGNOSIS OF EPIPHYSIAL SEPARATION

An Experimental Study

G. G. DALE and W. ROBERT HARRIS, TORONTO, ONTARIO, CANADA

From the Department of Anatomy, University of Toronto, and the Division of Orthopaedic Surgery, Toronto General Hospital

It has been clearly demonstrated that after experimental separation of an epiphysis the plane of separation is always on the diaphysial side of the epiphysial plate (Haas 1917, Harris 1950). The significance of this observation is that the growing cartilage cells in the plate remain adherent to the epiphysis. Thus, if the nutrition to these cells is not damaged by the separation, growth in the epiphysis should continue with little abnormality. This view is supported by several clinical studies (Aitken 1935*a*, 1935*b*, 1936; Aitken and Magill 1952, Aitken and Ingersoll 1956). Yet in a recently reported study of the histological changes after experimental separation of the upper femoral epiphysis, a different result was found (Harris and Hobson 1956). In this case the separation was followed by prompt avascular necrosis of the bony epiphysis and cessation of growth in the epiphysial plate. Repair occurred by new bone which grew across the remnants of the plate. It seems to us that this result might be peculiar to the upper femoral epiphysis, and that it could be related to the fact that, since the upper femoral epiphysis is entirely covered by articular cartilage, its nutrient vessels must enter it by being closely applied to the periphery of the epiphysial plate. In other words the vessels are so situated that they must inevitably be damaged in epiphysial separation. It seemed desirable, therefore, to extend our observations to include a study of the methods by which epiphyses are vascularised and the influence this has upon healing after epiphysial separation.

MATERIAL AND METHODS

The details of the blood supply to epiphyses were studied by means of India ink injections in M. Rhesus monkeys. (These animals were used for the production of Salk poliomyelitis vaccine and were provided through the courtesy of the Connaught Laboratories of the University of Toronto.) A cannula was inserted into the left ventricle, and the animal was perfused with a warm solution of 0·75 per cent sodium chloride, 0·5 per cent sodium oxalate and 0·2 per cent sodium nitrate, a mixture which combines vaso-dilating and anticoagulant properties. This preliminary perfusion was followed by an injection of a heated mixture of 30 cubic centimetres of India ink in 100 cubic centimetres of 5 per cent gelatin. When the circulatory system was completely filled with the latter solution the body of the animal was immersed in ice-water to set the gelatin. The epiphyses and adjacent soft tissues were dissected out, fixed in formalin, decalcified, cut into slabs about one millimetre thick, and finally cleared in oil of wintergreen by the Spaltehotlz technique (Bensley and Bensley 1938). The cleared specimens were then studied both with the dissecting microscope and by using the cleared slab as a " negative " and placing it in the carrier of a photographic enlarger to provide an enlarged photographic print.

RESULTS

Two basic forms of vascularisation were found; the type depended upon whether or not the epiphysis was entirely or only partly covered by articular cartilage (Fig. 1).

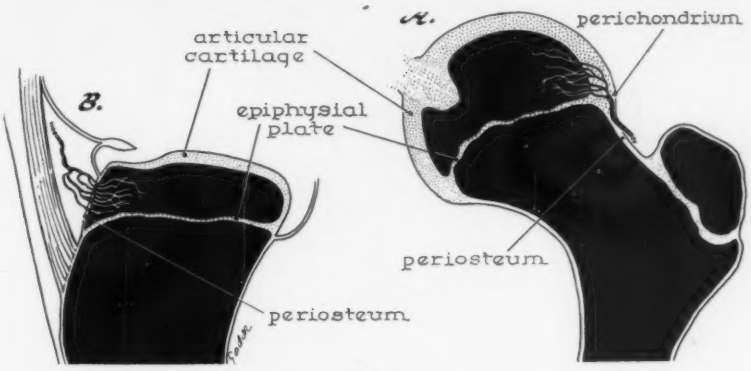

FIG. 1

Diagram to demonstrate that when an epiphysis is entirely covered by articular cartilage (*A*), its blood vessels must enter it by traversing the perichondrium at the periphery of the epiphysial plate. This makes them vulnerable to rupture after epiphysial displacement. By contrast when an epiphysis is only partly covered by articular cartilage (*B*) its blood vessels enter it in such a way that separation could occur without serious damage to them.

FIG. 2

FIG. 3

Figure 2—Cleared India ink injection of the femoral head of a monkey (M. Rhesus). The epiphysis is entirely covered by cartilage, and its nutrient vessels enter it by traversing the perichondrium at the periphery of the epiphysial plate. Figure 3—Cleared India ink injection of the upper tibial epiphysis of a monkey. The epiphysis is only partly covered by articular cartilage, and its nutrient vessel enters it by penetrating the cortical bone at the side of the epiphysis.

Type A. Epiphysis entirely covered by articular cartilage—Since vessels cannot traverse the joint space to enter this type of epiphysis, the nutrient vessel must enter the epiphysis by traversing the perichondrium at the periphery of the epiphysial plate* (Fig. 2). On entering the bony epiphysis proper, the main vessel breaks up into smaller branches. No direct communication through the epiphysial plate was observed between these vessels and those of the adjacent metaphysis.

Type B. Epiphysis partly covered by articular cartilage—The blood vessels of this type of epiphysis enter the bone by penetrating the cortex at the side of the epiphysis (Fig. 3), just as does the nutrient vessel to the shaft of a long bone. Often more than one vessel enters the epiphysis or a single vessel branches as it enters the epiphysis itself. Again, no direct communication through the epiphysial plate was seen between these vessels and those of the metaphysis.

The upper femoral and upper radial epiphyses are good examples of Type A epiphyses, while most of the rest of the epiphyses in the body may be classified as Type B.

DISCUSSION

The site of the nutrient vessels in relation to the epiphysial plate in each of the two types of epiphysis suggests very strongly that an epiphysis of Type B could be separated with little damage to its blood vessels, while separation of an epiphysis of type A would almost certainly produce serious vascular damage. Experimental proof of the latter hypothesis is provided by the study of the changes after experimental separation of the upper femoral epiphysis (Harris and Hobson 1956). Although there is ample clinical evidence to support our reasoning about the fate of the blood vessels after separation of a type B epiphysis, few experimental studies are available and these lack detail (Kühne 1952, Müller 1924). For this reason the remainder of our investigation was confined to a study of the histological changes found after separation of a type B epiphysis.

MATERIAL AND METHODS

Because of its accessibility and the ease with which it can be separated the distal radial epiphysis was selected as an example of a type B epiphysis. Four to six weeks old rabbits were used. The epiphysis was exposed subperiosteally through a longitudinal incision. It was easily separated by manual hyperextension of the forepaw. After reduction the periosteum and skin were closed. The shape of the epiphysial plate is such that the reduction was stable, so no fixation was required. The animals were killed at two-day intervals up to ten days, and thereafter at weekly intervals up to four weeks. There were ten animals in each group, making a total of eighty for the whole experiment. Both distal radial epiphyses were recovered, fixed and decalcified in Bouin's fluid and sectioned in paraffin.

RESULTS

Immediately after separation the gap on the diaphysial side of the epiphysial plate filled with fibrin (Fig. 4). Although occasional fragments of necrotic bone and cartilage were seen on the metaphysial side of the separation the metaphysis as a whole remained viable. However, the new bone formation responsible for endochondral ossification ceased. In consequence the calcified cartilage cells on the diaphysial side of the plate persisted unaltered. Simultaneously there was continued growth of the young cartilage cells on the epiphysial side of the plate.

* The ligamentum teres of the femoral head is an apparent exception to this rule. However, in our experience the artery of the ligamentum teres usually terminates at the insertion of that ligament to bone, without supplying a significant amount of the epiphysial bone itself.

These two processes occurring together caused a temporary but pronounced increase in the thickness of the plate (Fig. 5). Finally, callus began to grow from the metaphysis and the periosteum of the shaft across the gap produced by the separation, and united the epiphysis to the shaft (Fig. 6). Once this had occurred, normal endochondral ossification resumed and the thickness of the plate rapidly returned to normal (Fig. 7). At no time during the healing process was there any evidence of avascular necrosis in the bony epiphysis.

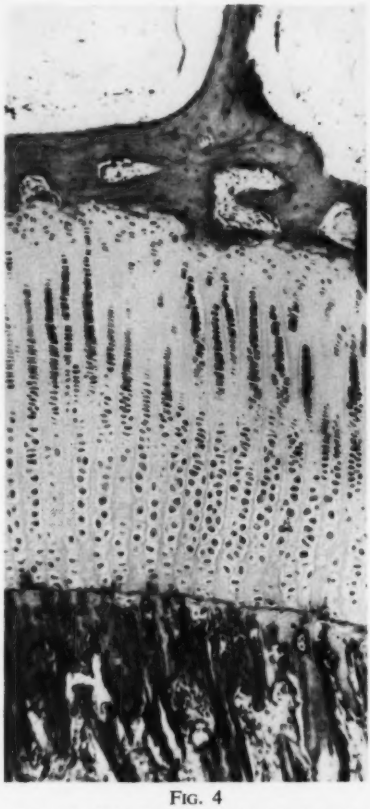

FIG. 4

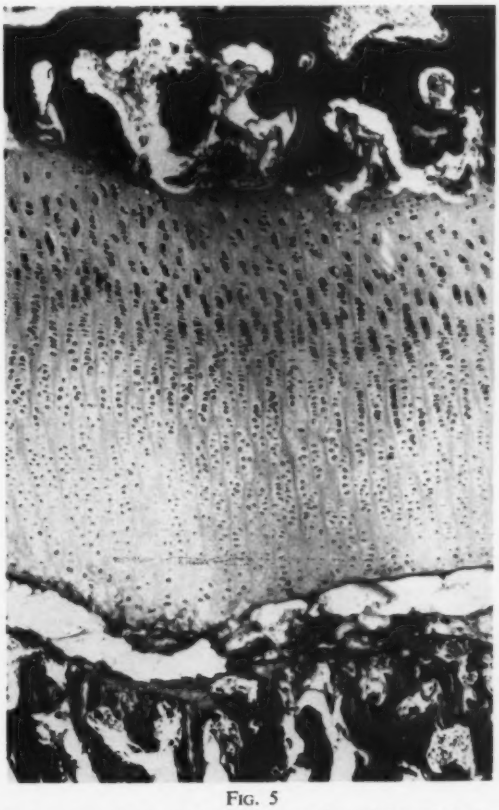

FIG. 5

Figure 4—Distal radial epiphysial plate of a rabbit, two days after experimental separation of the epiphysis. The narrow line of separation is filled with fibrin and can be seen on the diaphysial side of the plate.
Figure 5—Distal radial epiphysial plate of a rabbit ten days after separation. Note the tremendous increase in the thickness of the plate. This is brought about by the temporary cessation of endochondral ossification which allows the dead cartilage cells in the zone of provisional calcification to persist unaltered.

The time sequence in each case followed a regular pattern. The increased thickness of the epiphysial plate reached its maximum between seven and ten days, and endochondral ossification was fully re-established by two weeks. After three weeks it was virtually impossible to distinguish between the detached epiphysis and the untreated control epiphysis.

DISCUSSION

The results of this experiment demonstrate that an epiphysis of type B can be separated from its shaft without serious impairment of its capacity for growth. In view of the method by which this type of epiphysis receives its blood supply it seems fair to conclude that its nutrient vessels are not damaged by the separation. The striking contrast between these

results and those already mentioned after separation of the upper femoral epiphysis leads us to conclude that the prognosis of epiphysial separation is dependent primarily on the amount of damage to the epiphysial blood supply rather than the actual mechanical disturbance to the epiphysial plate. It is recognised that epiphysial plates may be damaged either by crushing or by being split open at right angles to their diameter so that a regional closure of the plate with subsequent deformation of growth occurs. This particular form of injury was not investigated in this study.

FIG. 6

Distal radial epiphysis in the rabbit two weeks after experimental separation. Endochondral ossification is resuming but the former line of separation can still be seen.

A coincidental finding of this experiment is that its results throw some light on the question of how epiphysial plates are nourished. Three possible pathways exist: from the epiphysis, from the metaphysis or from the perichondrium attached to the periphery of the epiphysial plate. It is commonly accepted that the principal route is from the metaphysis, and this view is supported by the recent studies of Brodin (1955). The present investigation suggests that the epiphysial cartilage cells in an epiphysis of type B can survive and indeed proliferate after complete separation from the metaphysis. It does not, however, determine

whether these cells secure their nourishment from the epiphysis proper or from the soft-tissue attachments at the periphery of the plate.

SUMMARY AND CONCLUSIONS

1. The methods by which epiphyses receive their blood supply was studied by means of India ink injections in monkeys. Two types were identified depending upon whether the epiphysis was entirely or partly covered by articular cartilage. In the former, nutrient vessels

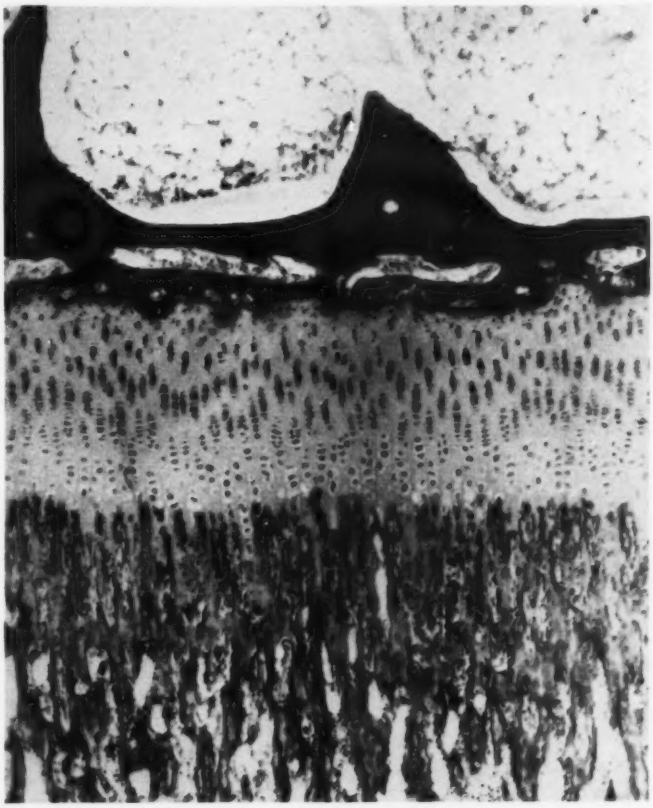

FIG. 7

Distal radial epiphysis in the rabbit twenty-four days after experimental separation. Endochondral ossification is fully restored, and both in size and appearance the plate is indistinguishable from the normal.

enter the epiphysis by traversing the perichondrium at the periphery of the plate. In the latter they enter the epiphysis by penetrating the cortex at the side of the epiphysis at a point remote from the epiphysial plate.

2. The histological changes after separation of the second type of epiphysis were also studied. After temporary interference with endochondral ossification marked by increased thickness of the epiphysial plate, healing occurred so rapidly that within three weeks it was difficult to determine that the epiphysis had been separated at all.

3. It is concluded that when nutrient vessels enter an epiphysis at a point remote from the epiphysial plate, that epiphysis can be separated without serious disturbance to its blood supply and accordingly without interference with its capacity for growth. As it has been established that an epiphysis which is entirely covered by articular cartilage cannot be separated without destruction of its blood supply and subsequent avascular necrosis (Harris and Hobson 1956), it is concluded that the prognosis of an epiphysial separation is dependent upon the degree of damage to its blood supply rather than the mechanical disturbance of the epiphysial plate.

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REFERENCES

AITKEN, A. P. (1935*a*): The End Results of the Fractured Distal Radial Epiphysis. Journal of Bone and Joint Surgery, **17,** 302.

AITKEN, A. P. (1935*b*): Further Observations on the Fractured Distal Radial Epiphysis. Journal of Bone and Joint Surgery, **17,** 922.

AITKEN, A. P. (1936): End Results of Fractures of the Proximal Humeral Epiphysis. Journal of Bone and Joint Surgery, **18,** 1,036.

AITKEN, A. P., and MAGILL, H. K. (1952): Fractures Involving the Distal Femoral Epiphyseal Cartilage. Journal of Bone and Joint Surgery, **34–A,** 96.

AITKEN, A. P., and INGERSOLL, R. E. (1956): Fractures of the Proximal Tibial Epiphysial Cartilage. Journal of Bone and Joint Surgery, **38–A,** 787.

BENSLEY, R. R., and BENSLEY, S. H. (1938): Handbook of Histological and Cytological Technique. Chicago: University of Chicago Press.

BRODIN, H. (1955): Longitudinal Bone Growth. The Nutrition of the Epiphyseal Cartilages and the Local Blood Supply. Acta Orthopaedica Scandinavica, Supplementum No. 20.

HAAS, S. L. (1917): The Localization of the Growing Point in the Epiphyseal Cartilage Plate of Bones. American Journal of Orthopedic Surgery, **15,** 563.

HARRIS, W. R. (1950): The Endocrine Basis for Slipping of the Upper Femoral Epiphysis. Journal of Bone and Joint Surgery, **32–B,** 5.

HARRIS, W. R., and HOBSON, K. W. (1956): Histological Changes in Experimentally Displaced Upper Femoral Epiphyses in Rabbits. Journal of Bone and Joint Surgery, **38–B,** 914.

KÜHNE, H. (1952): Zur Physiologie der Epiphyse des Knochens. Zentralblatt für Chirurgie, **77,** 2,193.

MÜLLER, W. (1924): Experimentelle Untersuchungen über Nekrosen und Umbauprozesse am Schenkelkopf nach traumatischen Epiphysenlösungen und Luxationen und ihre klinische Bedeutung. Bruns' Beiträge zur klinischen Chirurgie, **132,** 491.

OSTEOARTHRITIS OF THE HIP

An Electron Microscope Study*

K. LITTLE, LIONEL H. PIMM and J. TRUETA, OXFORD, ENGLAND

From the Nuffield Orthopaedic Centre, Oxford, and the Atomic Energy Research Establishment, Harwell

In their study of the nature and evolution of osteoarthritis of the hip Harrison, Schajowicz and Trueta (1953) showed that the initial lesion took place in the articular hyaline cartilage. They showed furthermore that the earliest sites of the degenerative changes were the non-pressure areas of the joint (Fig. 1). These workers and others stressed the importance of joint function in maintaining the health and nutrition of the articular cartilage (Trueta 1956,

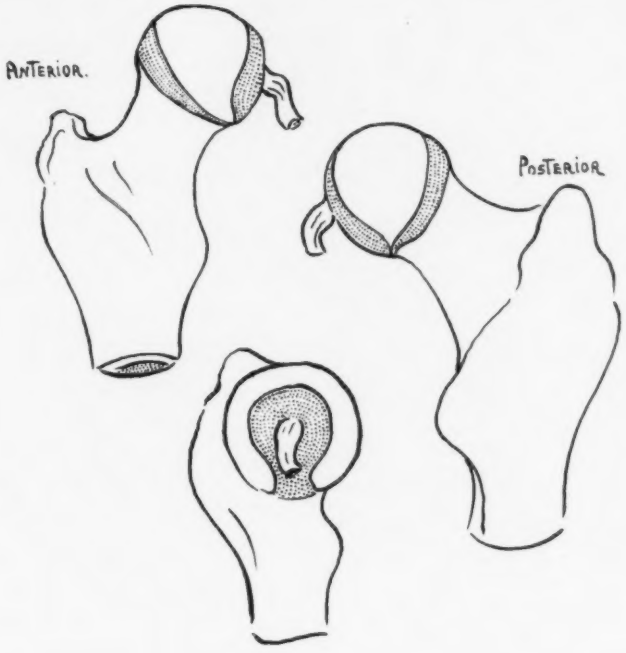

FIG. 1

The shaded areas represent the non-pressure areas of the joint, where degenerative changes first occur.

Ingelmark and Sääf 1948, Ingelmark 1950, Ekholm 1951, Ekholm and Norbäck 1951). It appears that function exerts its influence through the intermittent pumping action of alternate pressure and rest, as Müller (1929) suspected. This action has been likened by Trueta (1954) to that of a bath sponge. Matthews (1952) showed that the weight-bearing area comprises the thickest hyaline cartilage with the greatest content of mucopolysaccharide. It seems therefore that the whole key to osteoarthritis lies in the understanding of the earliest changes in the hyaline articular cartilage.

* Based upon a paper read by Mr L. H. Pimm to the First Regional European Conference on Electron Microscopy in 1956.

The histological changes that occur in the osteoarthritic joint have been studied in detail by many workers, and it is doubtful whether the light microscope will have much place in further elucidating the problem. The increased resolution of the electron microscope should, however, enable a more detailed study of the cartilage changes to be made. During the past three years a technique for such study has been evolved, and this paper records the preliminary results of these investigations.

METHOD OF STUDY

Techniques of electron microscopy—Because of the higher resolving power of the electron microscope, greater care is needed in fixing material for it than for the conventional light microscope, by which fixation artefacts often cannot be resolved. In the present study, specimens for embedding and sectioning were cut into cubes not more than two or three millimetres across and fixed in a variant of Palade's osmic solution, or occasionally in neutral

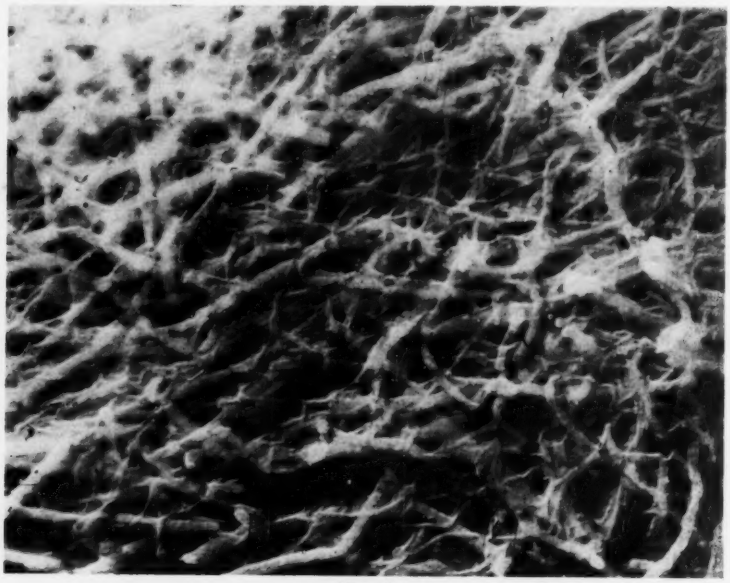

FIG. 2

Cartilage. Torn edge of section, to show sizes of collagen fibrils which are present in cartilage. From under medial surface of head of femur in a case of osteoarthritis of the hip. (×20,000.)

formalin. They were then dehydrated with absolute alcohol, and pieces of about a millimetre across were placed in a monomer containing 17 per cent isobutyl methacrylate in butyl methacrylate. When dry this can be polymerised in ultra-violet light, and the consistency of the block containing the specimen for sectioning may be controlled by varying the time and rate of polymerisation. This monomer and method of polymerisation were found to be more satisfactory than heating methyl methacrylate monomer, because the embedding medium could later be removed from sections without distorting the specimen. The block containing the specimen was mounted in a copper block on a microtome, which was cooled with solid carbon dioxide, the subsequent expansion of the copper providing a slow advance. Sections were cut with a glass knife.

Occasional thin sections were examined, but on the whole more information was obtained from sections about 0·1–0·3μ thick. The methacrylate was removed with amyl acetate, and stereoscopic photographs of the sections were taken with a Philips microscope with an

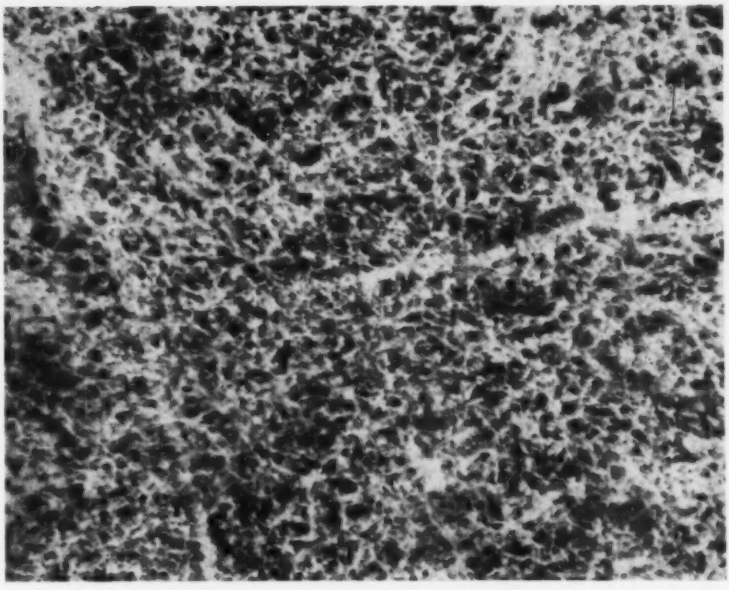

FIG. 3

Normal human articular cartilage, obtained at operation in a patient aged twenty-two. There is a random network of collagen fibrils of assorted sizes in the ground substance. This matrix appears as a dried gel, with spaces taking the place of fluid. (×10,000.)

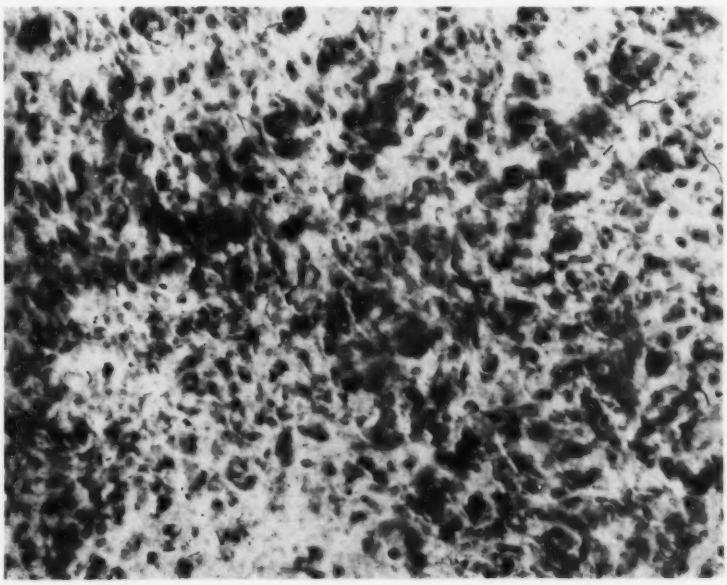

FIG. 4

Normal articular cartilage from the femoral condyle of a rabbit. Note the random three-dimensional network of collagen fibrils in the matrix of ground substance, which appears as dried gel. (×20,000.)

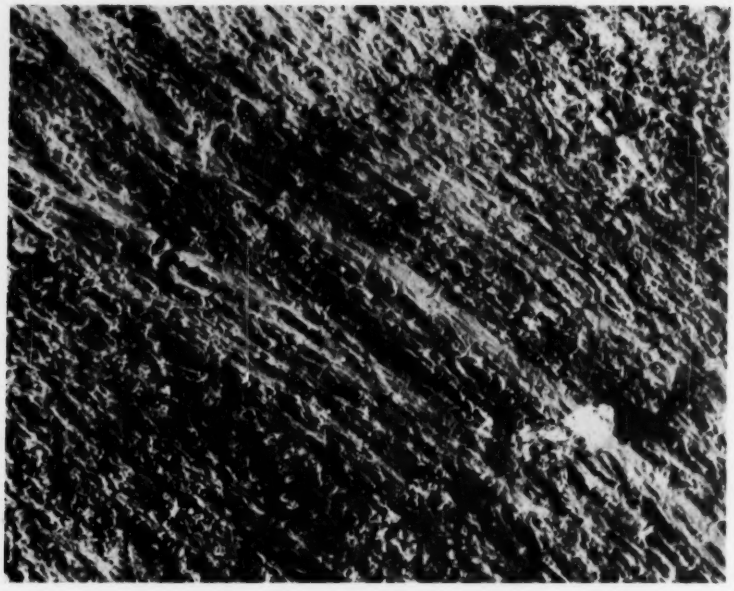

FIG. 5
Cartilage from femoral head in osteoarthritis of the hip. (×5,000.)

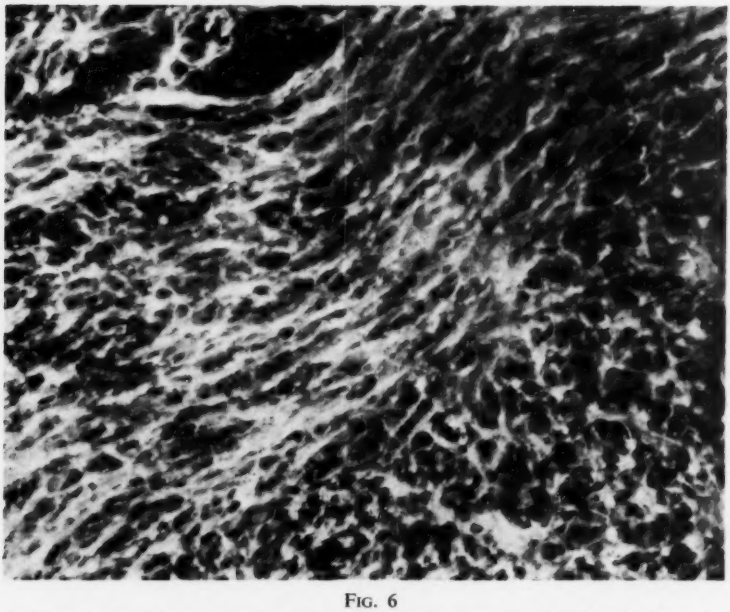

FIG. 6
Articular cartilage from upper part of head of femur in osteoarthritis of the hip. Note the orientation of the collagen fibrils, which comparison with x-ray diffraction photographs shows to be perpendicular to the surface of the bone. The collagen fibrils are less thickly covered with ground substance than in normal articular cartilage. (×20,000.)

FIG. 7
Cartilage from femoral head in osteoarthritis of the hip. (×5,000.)

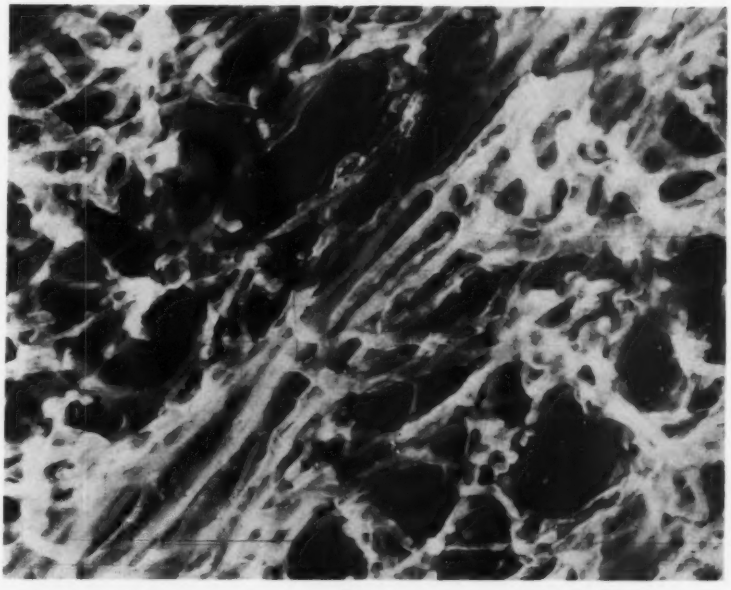

FIG. 8
Articular cartilage from upper part of head of femur removed at operation in a case of osteoarthritis of the hip. The orientation is again very marked, and with a lower proportion of ground substance. (×40,000.)

objective lens capable of resolving 20 Å. The stereoscopic technique was found to give a better idea of the interrelation of the components. Some sections were shadowed: this had the effect of making the section more stable in the electron beam, and producing more beautiful pictures, but more information could be obtained from unshadowed sections, because shadowing affects only one surface of a section and tends to mask detail underneath.

X-ray diffraction techniques—To find the orientation of collagen over larger fields than can conveniently be covered in the electron microscope x-ray diffraction was used. Specimens were fixed in formalin, and strips about 0·25–0·5 millimetre thick were cut in the required plane. Diffraction photographs were taken using Cu Kα radiation and a flat plate fibre camera with the plate three centimetres from the specimen. The x-ray beam covered an area of the specimen about two-thirds of a millimetre in diameter. The specimen could be held at

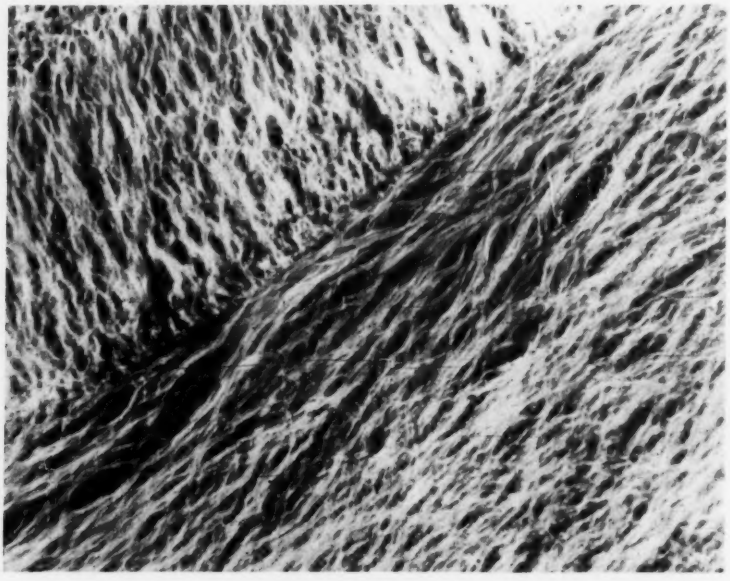

FIG. 9

Fibrocartilage from knee. The collagen is arranged in bundles, which tend to be randomly orientated. The photograph shows a junction between two bundles. (×10,000.)

any desired angle. From the orientation of the collagen diffraction pattern on the plate and the known geometry of the specimen, the orientation of collagen in the specimen could be deduced.

RESULTS

With the technique described, normal hyaline cartilage viewed in the electron microscope appears to consist of collagen fibrils closely surrounded by a gelatinous or spongy matrix containing polysaccharide. Four distinct sizes of collagen fibril have been noted (Fig. 2) and these fibrils are normally present in a three-dimensional network with no preferred orientation (Figs. 3 and 4). In the hyaline cartilage from the arthritic joint two differences are apparent. First the collagen has a definite orientation (Figs. 5 and 6). Secondly less ground substance is shown, so that the material has a rather more open appearance (Figs. 7 and 8). This is in agreement with the chemical analyses performed by Matthews (1952), in which it was found that the total quantity of polysaccharide was decreased considerably in arthritic cartilage.

Although the onset of the changes in orientation can be seen easily in sections viewed in the electron microscope, the area of specimen examined, and indeed the whole specimen, is so small that it is difficult to find the direction of this orientation. Information on this point can be gained by using x-ray diffraction in conjunction with electron microscopy. A survey

FIG. 10

FIG. 11

FIG. 12

Figure 10—Flat plate x-ray diffraction pattern of normal cartilage. The sharp rings are due to collagen, and some of the more diffuse background is caused by the ground substance. Lack of orientation can be deduced because the collagen diffraction pattern is in the form of complete rings. Figure 11—Flat plate x-ray diffraction pattern of ageing cartilage. Contraction of the collagen reflections into arcs indicates that it is orientated. From the orientation of this diffraction pattern on the plate and the known geometry of the specimen, the orientation of the collagen in the specimen can be deduced. Figure 12—Flat plate x-ray diffraction pattern of arthritic cartilage. The orientation of collagen fibrils, as shown by the sharper arcs, is greater than in normally aged cartilage. Less diffuse background scattering suggests a lower proportion of ground substance.

of the changes in orientation produced both by normal ageing and by the onset of osteoarthritic changes has therefore been carried out using x-ray diffraction.

In the foetus and newborn child diffraction shows that the whole of the cartilage is unorientated except for the immediate surface layer, in which the collagen fibrils lie parallel to the surface. (The diffraction pattern resembles that in Figure 10.) This lack of orientation could be due to one of two causes: either, as in the present case, the collagen fibrils themselves are

randomly orientated; or, as in fibrocartilage, such as the semilunar cartilages of the knee (Fig. 9), the bundles of collagen are randomly orientated. It requires the electron microscope to distinguish between these two.

In normal ageing, by the time middle age is reached the orientation on the surface is still parallel with that surface, with a tendency towards preferred orientation away from the ligamentum teres. The outer two-thirds or so of the articular cartilage remains unorientated (Fig. 10), but there is a tendency for the inner layers of cartilage adjacent to the bone to show definite orientation at right angles to the surface (Fig. 11).

In osteoarthritic hips the entire thickness of the cartilage has its collagen fibrils orientated at right angles to the surface of the bone, with the exception still of the fibrils in the surface

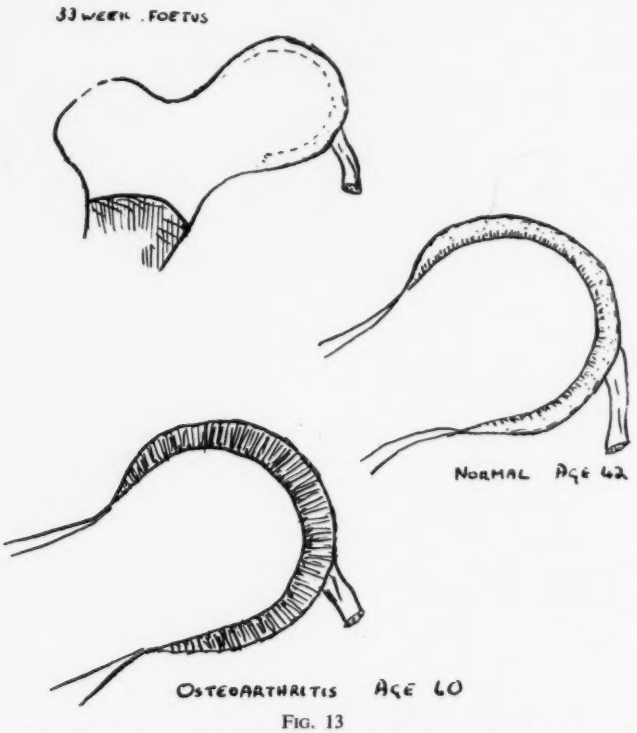

FIG. 13

Diagram of section through head of femur, to illustrate changes in orientation that occur in normal ageing and in osteoarthritis.

layer of the cartilage, which remain parallel to the surface till the cartilage has begun to split and wear away (Fig. 12). It seems that this change in the direction of the collagen fibrils is most easily explained in terms of tensional forces across the joint, which is in agreement with the observation that osteoarthritis first appears in the non-weight-bearing areas. These changes in orientation are summarised in Figure 13.

Living and dead cartilage cells have been encountered in both normal and arthritic cartilage, with possibly more in the latter. In dead cells the cytoplasm is replaced by fibrous tissue.

Although this study is at present in its earliest stages we believe that electron microscopy combined with x-ray diffraction will further our knowledge of the structure of articular

cartilage, for we have shown that definite changes in this structure occur in degenerative arthritis. The full significance of these changes is now being investigated.

SUMMARY

1. A study of normal and osteoarthritic hyaline cartilage has been made with the electron microscope and x-ray diffraction.
2. Normal cartilage consists of a three-dimensional network of collagen fibrils with no preferred orientation, surrounded by a matrix containing polysaccharide.
3. In the osteoarthritic joint the collagen fibrils show definite orientation and a decreased proportion of ground substance. X-ray diffraction confirms this and shows the orientation to be at right angles to the surface of the femoral head.
4. Tensional forces across the joint may explain why osteoarthritic changes first appear in the non-weight-bearing area of the joint.

REFERENCES

EKHOLM, R. (1951): Articular Cartilage Nutrition. How Radioactive Gold Reaches the Cartilage in Rabbit Knee Joints. Acta Anatomica, Supplementum **15.**

EKHOLM, R., and NORBÄCK, B. (1951): On the Relationship Between Articular Changes and Function. Acta Orthopaedica Scandinavica, **21,** 81.

HARRISON, M. H. M., SCHAJOWICZ, F., and TRUETA, J. (1953): Osteoarthritis of the Hip: A Study of the Nature and Evolution of the Disease. Journal of Bone and Joint Surgery, **35–B,** 598.

INGELMARK, Bø. E. (1950): The Nutritive Supply and Nutritional Value of Synovial Fluid. Acta Orthopaedica Scandinavica, **20,** 144.

INGELMARK, Bø. E., and SÄÄF, J. (1948): Über die Ernährung des Gelenkknorpels und die Bildung der Gelenkflüssigkeit unter verschiedenen funktionellen Verhältnissen. Acta Orthopaedica Scandinavica, **17,** 303.

MATTHEWS, B. F. (1952): Collagen/Chondroitin Sulphate Ratio of Human Articular Cartilage Related to Function. British Medical Journal, **ii,** 1,295.

MÜLLER, W. (1929): Biologie der Gelenke. Leipzig: Johann Ambrosius Barth.

TRUETA, J. (1954): Osteoarthritis of the Hip. Annals of the Royal College of Surgeons of England, **15,** 174.

TRUETA, J. (1956): Osteo-arthritis. An Approach to Surgical Treatment. Lancet, **i,** 585.

THE HYDRATION OF THE NUCLEUS PULPOSUS AND ITS RELATION TO INTERVERTEBRAL DISC DERANGEMENT

NEIL G. C. HENDRY, ABERDEEN, SCOTLAND

The natural history of the normal disc is well established. From the time of the earliest investigations (Uebermuth 1929, Puschel 1930, Junghanns 1931, von Puky 1937) it has been clear that the efficient functioning of the disc depends largely on the elasticity of the nucleus pulposus, and that this in turn is closely related to its water-binding capacity. In early life a water content of 80–88 per cent is usually quoted. From the fourth decade onwards an increasing proportion of subjects show a progressive lowering of the degree of hydration until, in late life, figures of 70 per cent may occur (Keyes and Compere 1932, Lindahl 1948). This change is usually accompanied by an alteration in the appearance of the nucleus, which becomes amorphous and sometimes discoloured, and then progressively more and more fibrotic until all trace of the normal highly elastic and highly hydrated gel is lost. Sylvén (1951) showed that the nucleus consists of a three-dimensional lattice of collagen fibrils in which is enmeshed a muco-protein gel. This protein-polysaccharide complex is responsible for the hydrophilia of the nucleus, and suffers a patchy loss, and disappearance of gel structure, as age advances. Large collagen bundles are laid down, with consequent lowering of the water content, until eventually, in advanced degeneration, the collagen may be completely devoid of mucoid material. The increased deposition of collagen, and possibly of elastin also, has been confirmed by other writers (Naylor, Happey and McRae 1954). This is therefore undoubtedly the abnormality that underlies and explains the dehydration occurring in the nucleus in later life. Although in general it occurs from the fourth decade onwards, it is not universal, and the persistence of a high water content into the sixth decade has been reported (Coventry, Ghormley and Kernohan 1945).

There is therefore a generally accepted concept of the process of " ageing " of the normal intervertebral disc, and considerable and growing understanding of the biochemical changes responsible for its gradually decreasing efficiency. In contrast, alterations in the behaviour and properties of protruded or extruded discs have received little attention. Deuchar and Love (1939) reported the microscopical appearances of 100 deranged discs removed at operation. They found degeneration of at least slight or moderate degree in almost every case, irrespective of age, and advanced changes in some cases in all age groups. These changes were " not infrequent " in specimens from young persons, and their occurrence rate increased only slightly with advancing age. Eckert and Decker (1947) published similar results. In forty " normal " discs from cadavers of all ages degenerative changes were not found in the first three decades, but had a gradually increasing incidence thereafter. In 182 mechanically deranged discs, on the other hand, changes were found irrespective of age, including surprisingly large areas of degeneration in material from several patients in the second decade. Such evidence as is available, therefore, suggests that, whether before or after displacement, deranged discs suffer either an acceleration of the normal ageing process or the action of some other factor which the normal disc escapes.

This paper pursues the same question by comparing the water-binding capacity (and therefore the functional efficiency) of normal discs obtained at necropsy with that of mechanically deranged discs obtained at laminectomy.

PHYSICO-CHEMICAL BASIS OF WATER RETENTION WITHIN THE NUCLEUS

An essential preliminary to any investigation of this sort is an understanding of the mechanism whereby water is retained in the disc. There are two possibilities: osmotic pressure exerted by the individual molecules; and imbibition pressure exerted by the protein/polysaccharide gel. In general, the relation of these forces one to another is a complicated one, and I know of little published evidence concerning their relative importance in the disc. There has, however, been an almost universal tendency to assume that osmosis is the only factor, or, at least, the only important one. Inman and Saunders (1947) stated that the disc is to be regarded as an osmotic system in which the delicate mechanism of water balance can be upset by even minute tears in the annulus or the plates, with consequent interference with their function as semi-permeable membranes. The desiccation of the nucleus with advancing age is ascribed to decreasing permeability of the cartilage end-plates. Birkett (1950) summarised the published opinion up to that date and concurred, although less dogmatically, with the same view. Naylor and Smare (1953) suggested that the breakdown of complex molecules within the nucleus to produce simpler but more numerous ones would increase osmotic attraction, increase the pressure within the disc, and consequently cause or predispose to rupture of the annulus. A similar " hyper-hydration " mechanism had already been postulated by Charnley (1952), but with the reservation that he did not think that osmosis could be responsible for it. Bush, Horton, Smare and Naylor (1956) suggested that the water content was regulated by the balance obtaining between osmotic pressure differences on the one hand and the cohesive forces of the tissue on the other.

There are many difficulties in the way of accepting osmosis as the source of the hydrophilia of the nucleus pulposus. There is no doubt that molecular breakdown in an osmotic system would, as postulated by Naylor and Smare, result in increased osmotic pressure and increased hydration. In degeneration of the nucleus, however, the reverse effect has universally been observed, and degeneration and dehydration have consistently been found to proceed hand in hand. In longstanding osteoporosis of the spine and in some spinal fractures the vertebral end-plates may be ruptured and no longer semi-permeable, yet ballooning of the neighbouring space occurs and may be progressive. The narrowing of disc spaces after accidental damage to the annulus at lumbar puncture has been attributed to the loss of semi-permeability (Pease 1935, Haas 1939) but, as Milward and Grout (1936) pointed out in recording the same phenomenon, the time taken for changes to appear is too great to be explicable on the basis of water loss. In an osmotic system, dehydration could occur only as a result of changes in the semi-permeable membrane, yet gross alterations are commonly found in the nucleus of discs examined post-mortem, whereas cartilage plate defects are rare (Saunders and Inman 1940). Coventry, Ghormley and Kernohan (*loc. cit.*), in describing discs studied at necropsy, specifically referred to some in which the bulk and the water content of the nucleus were decreased without any abnormality being present in the annulus or end-plate. Finally, it seems most unlikely on teleological grounds that so heavily stressed a structure, whose function depended upon the maintenance of an intact osmotic system, would consist predominantly of extra-cellular material, for the whole nucleus would then be rendered ineffective by the smallest defect in the containing membrane.

When Charnley (*loc. cit.*) expressed doubt about the part played by osmosis, he supported his statement with the observation that disc substance immersed in four-times-normal saline increased in weight almost to the same extent as when immersed in normal saline. This finding in itself would serve almost completely to discount osmotic attraction as an effective force in disc hydration, but confirmation and some slight elaboration seemed desirable. Eight pieces of nucleus from young normal post-mortem spines were immersed successively for twenty-four hours in twice-normal, normal and half-normal saline, and then in water, and weighed after each immersion. Pieces of annulus of approximately the same size and shape from the same discs were similarly treated. In every case the annulus showed the

successive increases in weight that were to be expected on the basis of an osmotically controlled hydration: in no case did the nucleus do so. There was little difference in the pattern of weight change between individual discs, and a typical result is shown in Figure 1. The maximum weight for annulus and nucleus is expressed as 100, and the other weights as a percentage. It will be seen that this nucleus was able to maintain an almost constant degree of hydration in the face of changes in tonicity which caused a major alteration in the hydration of its annulus. This difference in behaviour can be attributed only to the muco-protein gel of the nucleus, and makes it virtually certain that the hydration of the nucleus is predominantly due to the imbibition pressure of the gel.

The remainder of the work to be described is based on the assumption that measurement of the imbibition pressure of a nucleus provides an accurate index of its efficiency.

THE NATURE OF IMBIBITION PRESSURE AND ITS MEASUREMENT

Standard text-books of colloid chemistry give full descriptions of imbibition pressure and emphasise its complete independence from osmosis. The short summary that follows has been compiled from Gortner (1938) and McBain (1950).

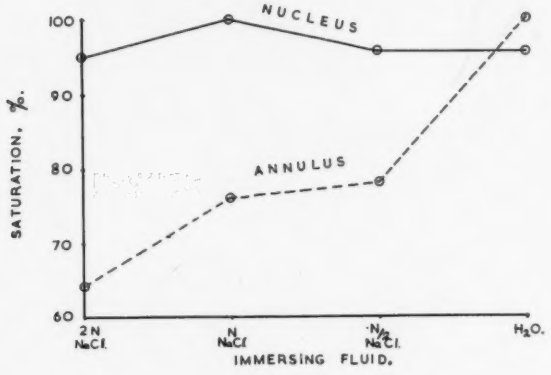

FIG. 1

Graphs illustrating the insusceptibility of the nucleus pulposus to changes in tonicity. The difference in behaviour between nucleus and annulus is to be attributed to the imbibition pressure of the nuclear gel.

Gels have two constituents, a solid or " disperse " phase (the protein/polysaccharide complex in the case of the nucleus pulposus) and a " dispersions medium " (in this case tissue fluid). The affinity between the two constituents causes them to exhibit the phenomenon known as hydration, solvation or imbibition, and is such that a pressure, known as the imbibition pressure, is required to force them apart. If the gel is less than fully hydrated it will absorb its specific dispersions medium against this same pressure. The magnitude of the pressure varies inversely with the degree of saturation, being high when the gel is poorly hydrated and falling to zero when solvation becomes complete. Although there is no possibility of major degrees of dehydration ever occurring in a gel in the human body during life, it is interesting to note the enormous forces that can be brought into play by the solvation mechanism. A gel with a saturation of 1 per cent will have an imbibition pressure of the order of 5,000 atmospheres, and in complete dehydration of the disperse phase the pressure is theoretically infinite. It is this force which is responsible for the swelling of slippery elm, for the extraction of water from saturated salt solutions by dried gelatine, for the continued hydration of the cactus in spite of the osmotic pressure exerted by dry salty soil, for the

ability of mushrooms to lift paving stones and dried peas to split rocks, and for many other familiar natural phenomena.

It is clear that this characteristic of gels provides a powerful and infinitely adaptable force to meet the functional needs of the intervertebral disc. It is indeed doubtful if any other mechanism could provide the strength, resilience and flexibility that are required.

Imbibition can be measured either by the weight increase occurring, or by the pressure developed, on immersion of the gel in its specific dispersions medium. Because of the progressive change in imbibition pressure as hydration proceeds, however, the initial degree of solvation must be known before any significance whatever can be attached to the result. Thus the isolated observation that a nucleus pulposus increases in weight or builds up a pressure on contact with saline or water indicates only that it was originally less than fully hydrated; it provides no information on the actual imbibition capacity of the nucleus, and no basis for the comparison of one nucleus with another. Failure to appreciate this has resulted, as will be seen later, in at least one widespread misconception concerning the hydrophilic properties of prolapsed discs.

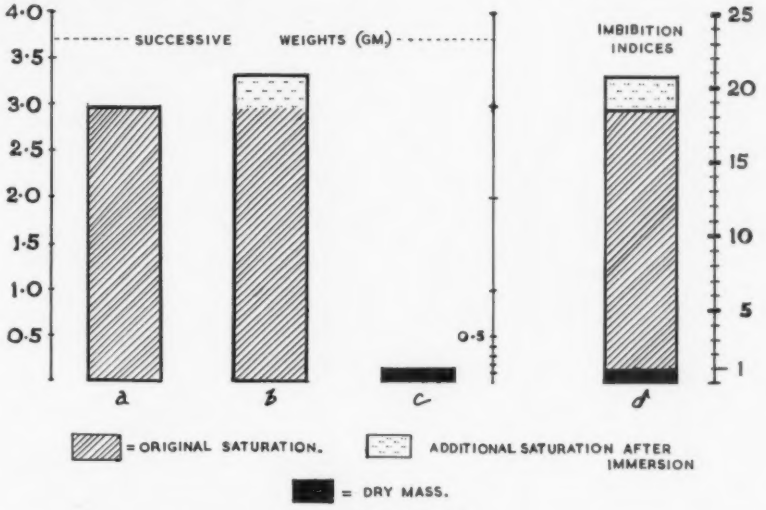

FIG. 2

To illustrate the relation between the successive weights of a specimen of disc material and the "imbibition indices" which are suggested as the best means of comparing one disc with another. The original weight (*a*), the fully hydrated weight (*b*) and the dry weight (*c*) combine to provide at (*d*) a representation of the hydrophilic power of one unit of dry weight of disc material.

COMPARISON OF THE IMBIBITION PRESSURES OF NORMAL AND DERANGED DISCS

In this investigation weight differences and the direct estimation of the ratio of pressure to degree of saturation were both employed. In each case the degree of saturation was determined by calculation from the dry weight of each nucleus. In order to avoid inaccuracies from de-naturing of the muco-protein in the course of drying, desiccation was carried out after all other estimations were complete.

ESTIMATION OF IMBIBITION BY WEIGHT INCREASE

Eighteen normal discs obtained post-mortem and seventy-five specimens from laminectomy were compared. The normals were taken from subjects between the ages of sixteen and thirty-five and were removed by section *en bloc* through the fourth lumbar vertebral body, the first

piece of the sacrum and the appropriate pedicles. The nuclei of the two lowest discs were thus available for investigation with the least possible exposure to air. A piece of each was excised, comparable in mass to the average laminectomy specimen, and cross-hatched to produce a surface of similar extent. Any disc showing evidence of a protrusion, or of discoloration or other naked-eye abnormality of the nucleus, was discarded. Six of the eighteen post-mortem discs, although normal in appearance, behaved in such a way as to suggest that they might more properly fall into a transitional group (Fig. 3). As the exact dividing line between normal and abnormal is, however, uncertain, it seemed best to continue to accept the naked-eye criteria and these six discs were included in the normal series. Similar imbibition characteristics were found in three other discs obtained post-mortem, which were excluded from the series on the grounds of discoloration and an abnormally granular appearance. It seems likely, therefore, that the microscopic appearances of such discs should be investigated in the future and might profitably be correlated with their imbibition capacity. The operation specimens were obtained from seventy-two patients aged between seventeen and forty-four; they were consecutive except for the exclusion of two which appeared to be entirely cartilaginous and therefore not comparable with the necropsy material. The indications

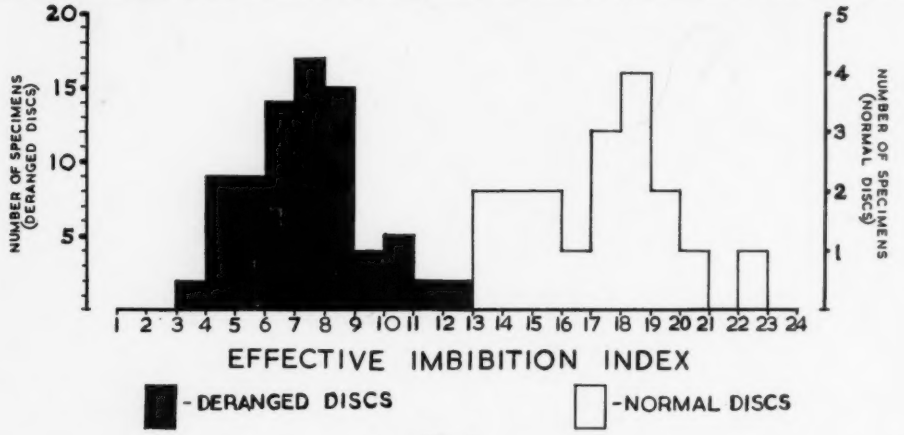

FIG. 3

To show the distribution of the effective imbibition index in seventy-five deranged and in eighteen normal discs. The six specimens whose indices fall between 13 and 16 may possibly form a transitional group.

for laminectomy had been the usual ones of failure to respond to, or early relapse after, adequate conservative treatment, and all derangements were therefore in an acute or " active " phase. At operation the disc substance was removed by pituitary rongeur or curette; the several fragments were collected on a gauze mop and then transferred to an airtight container to await weighing.

Each specimen, whether from operation or necropsy, was weighed, first in its fresh state, then after immersion in normal saline for twenty-four hours, and then after desiccation over anhydrous calcium chloride under reduced pressure. A comparison of the desiccated with the fully hydrated weight showed the maximum degree of saturation of which the nucleus was capable when free from pressure; and a comparison of the desiccated with the original weight indicated the degree of saturation which the nucleus had been able to maintain under the pressure obtaining in the disc space, or in the neural canal. The most convenient method of expressing these ratios, and of using them to compare one nucleus with another, is to take the desiccated weight in each case as unity and the other weights in proportion. Reference to Figure 2 will make this clear. This shows a representative normal disc, in which the excised portion of nucleus originally weighed 2·95 grammes. The weights after immersion and after

desiccation were respectively 3·32 grammes and 0·16 gramme. When the dry weight is taken as unity the original weight becomes 18·44 units and the fully hydrated weight 20·75. Figure 2 (*d*) therefore provides a representation of the hydrophilia of this nucleus, it being such that one unit of its dry substance could retain, in the conditions obtaining in the disc space, enough fluid to bring its weight to 18·44 units, and enough additional fluid, under conditions of free absorption, to increase its weight further to 20·75 units. Although the terms do not have general currency in the standard descriptions of the properties of gels, I suggest that, for the purpose of providing an accurate and easily determined means of comparing discs, the figures just quoted might properly be designated the " effective imbibition index " and the " free imbibition index " respectively. In so far as any one easily determined figure can do so, the former provides the most satisfactory measurement of the capacity of a nucleus to maintain its hydration, and therefore its efficiency, under its ordinary working circumstances. A third figure is important as indicating the reaction of the nucleus to pressure, or other factors in its environment, and that is the difference between the effective and the free indices. This might be called the "imbibition differential," and it will be noted that a high value for this would indicate an undue susceptibility to outside influences—that is, a lowering of the efficiency of the water-binding mechanism.

It will be seen that the apparent water content of the nucleus, as determined above, is higher than that usually quoted. The discrepancy is probably to be accounted for by the escape of some free gel into the surrounding saline, which, during the period of immersion, always became opalescent. Some loss no doubt also occurred during the removal of surface moisture after immersion. These losses are unfortunately unavoidable if desiccation is postponed until other observations are complete, as is essential. They appear likely to affect all discs equally, and not to prejudice the comparison of one with another.

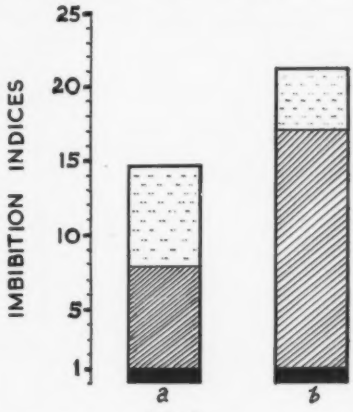

FIG. 4

A comparison of the imbibition characteristics of the " average " deranged disc (*a*) and the " average " normal disc (*b*). Same conventions as in Figure 2.

The imbibition characteristics of the normal (necropsy) and the deranged (operation) specimens were found to be markedly and uniformly different. As has already been shown, the effective imbibition index provides a measure of the water-binding capacity of the disc under what are probably its least arduous working conditions. In the normal group these indices lay between 13·3 and 22·4, with a mean of 17·1; whereas for the operation group the comparable figures were 3·4 to 12·7, with a mean of 7·9. The distribution throughout the two groups is shown in Figure 3, which makes clear the wide divergence between them. A less marked difference was found in the free indices, with mean figures of 21·2 (normal discs) and 14·7 (operation specimens). The " imbibition differential " was lower in the necropsy series (mean 4·1) and higher (mean 6·8) in the operative specimens. In this latter group individual differences were considerable and will be discussed in a later section. In spite of this variation, however, it is possible to give a reasonably accurate diagrammatic representation of the hydrophilic powers of an " average " prolapsed disc (Fig. 4). It will be seen that, even under conditions of free absorption, the deranged nucleus could fix much less fluid than the normal; and that under the conditions obtaining within the body, its water-binding capacity was even more markedly reduced. Its susceptibility to outside influences was greater in that a marked weight increase occurred on changing from bodily conditions to those of unhindered absorption. The significance of these findings is discussed below.

ESTIMATION OF IMBIBITION BY CORRELATING PRESSURE WITH THE DEGREE OF SATURATION

There are considerable technical difficulties in the way of obtaining a direct measurement of imbibition pressure in a heterogeneous structure such as a nucleus pulposus. Experiments with a hollow-bore needle communicating with the nucleus, either within the intervertebral space or in a rigid container, produced inconsistent results, and were not pursued. Fairly reliable figures could be produced at low loadings by simply placing the disc material in a syringe, weighting the piston and reading the volume changes on the syringe graduations. At even moderate pressures, however, jamming always occurred from the disc's insinuating itself between the piston and the barrel. An elaboration of this method proved fairly satisfactory—though occasionally somewhat temperamental—and reliable figures were eventually obtained for six normal and six protruded or extruded discs.

The weighed disc material was placed in a test tube with a perforated base; fine wire mesh, covered by a layer of filter paper, prevented extrusion through the perforations. The

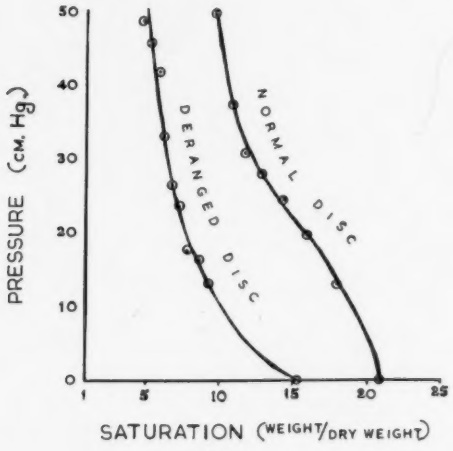

FIG. 5
Imbibition pressure saturation curves of representative normal and deranged discs.

remainder of the tube was occupied by a balloon, filled with a watery solution of gentian violet and retained in place by a rigid clamp. The balloon was connected, through a graduated glass tube, to a mercury manometer, and the meniscus between mercury and gentian violet was adjusted to correspond with the lowest graduation in the tube when the pressure in the balloon was zero. With the test tube and its contained disc immersed in normal saline, increasing pressure was applied step by step to the disc, and the successive resulting changes in its volume were noted by the movement of the mercury/gentian violet meniscus. The pressure that could usefully be employed was limited by the difficulty of preventing extrusion of the balloon at the upper end of the test tube—that is, of ensuring that all recorded changes of volume took place only in the disc. Usually 50 centimetres of mercury was the approximate limit, but considerably higher pressures were reached once or twice. When the limit of pressure had been reached and the final volume reading obtained the disc material was removed and weighed. A comparison of its actual weight with that calculated from the volume changes provided a rigid check on any experimental errors. The disc was then desiccated as before and weighed again.

The figures so obtained formed a progression in which the pressure contributed by the disc could be related to its saturation at each successive step. Figure 5 is a graph showing the pressure/saturation curves of one cadaveric and one operation disc. The saturations are expressed by the ratio of weight : dry weight, and are therefore exactly comparable with the imbibition indices described above. The saturation shown at 0 centimetre Hg. is, of course, the " free imbibition index."

The results yielded by this method, which in general conformed to the pattern shown in Figure 4, confirm those obtained by the simpler and probably more reliable weight-increase method, and add something to them. The discs obtained at operation were again markedly less hydrophilic than the normal, and they showed, particularly at low loadings, a greater than normal susceptibility to changes in pressure. It would be wrong to elaborate much on the results of so few cases, but it is of interest that, at a saturation corresponding to an imbibition index of 10, the pressures contributed are 43 centimetres Hg. for the normal disc and only 11 centimetres Hg. for the operation specimen. Both these specimens came from subjects aged thirty.

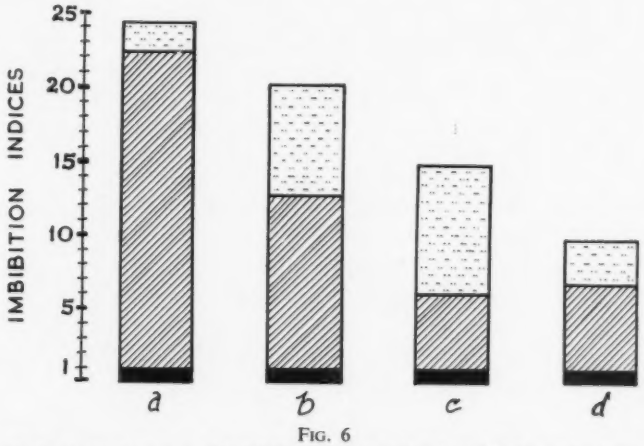

FIG. 6

To illustrate the range of imbibition characteristics found in the series from the most hydrophilic (necropsy) specimen (*a*) to the least hydrophilic (operation) specimen (*d*).

RANGE OF VARIATION IN IMBIBITION CHARACTERISTICS, AND CORRELATION WITH THE CLINICAL HISTORY

The imbibition indices of discs removed at operation showed a fairly wide variation within the limits which have been described. In general, the various patterns of behaviour were such as to support the conception of a gradually progressive change, corresponding to the duration of the derangement. Without there being any sharp dividing line between them, three phases could be distinguished, and the correlation between these and the length of the clinical history was often surprisingly exact. This was particularly so in patients with a long history of continuous disability, and also, although to a lesser extent, in those with symptoms of recent onset. The intermediate phase often corresponded with a recurrent disability, but the intermittent nature of the history in that event made exact correlation difficult.

What is thought to be the earliest phase of this progressive abnormality is shown in Figure 6 (*b*). This represents the most hydrophilic specimen of the discs obtained at operation and came from a man of thirty-eight with a history of only two months of acute sciatic pain. Operation was necessitated by paresis of the antero-lateral group of muscles in the affected

leg, advancing in spite of strict bed rest. Two completely separated portions of disc material were found, underlying an attenuated tensely bulging annulus, and weighing, together with a small quantity of curettings from the disc space, 4·83 grammes. The imbibition indices were 12·70 and 20·11. There were fifteen other specimens from patients whose histories extended over four months or less, and of these, eleven corresponded closely in type with the one illustrated—that is to say, they had a relatively high effective imbibition index and a high imbibition differential. A nucleus of this sort was capable, therefore, of retaining a normal or near normal amount of fluid when unstressed, but could not do so under the conditions obtaining in the intervertebral space; it seems reasonable to deduce from this that the gel was only moderately reduced in amount but was considerably reduced in efficiency. A similar imbibition pattern was found in two post-mortem discs, from subjects aged thirty-one to forty-four, which have already been mentioned and which were not included in the normal series because of brown discoloration of the nucleus.

At the opposite end of the scale of abnormalities were eight specimens of the type shown in Figure 6 (*d*), characterised by an effective index which was again rather higher than the average for deranged discs, but associated with a low imbibition differential. There was an absolute correlation between this imbibition pattern and a long clinical history, this group comprising all the six patients who had had a continuous disability for three years or more. Three patients were undergoing their second exploration at the same level, two of them after six years and one after ten. The specimen illustrated came from a man of fifty-two, who had a ten years' history of unremitting backache and sciatica. He had a large protrusion of the fourth lumbar disc, adherent to the fifth lumbar root, and yielding 5·62 grammes of material. These discs, in virtue of the very marked lowering of their capacity for free absorption, had an insusceptibility to pressure effects almost similar to that enjoyed by the normal disc at a high level of hydration. I suggest that this is probably to be explained by the gradual replacement of the gel with collagen, and that the next stage in the progression is one of fibrosis, hydrostatic inertness and clinical healing. This is to some extent borne out by the three specimens from second explorations, in which fibrosis could be expected to be most advanced. In these, the effective indices were 7·6, 7·8 and 7·9, with imbibition differentials of only 2·4, 2·3 and 2·6 respectively.

The remaining fifty-one discs obtained at operation occupied a position between the two extremes just described. It was in this group that the lowest effective indices were found, and the free indices were intermediate, ranging between 10 and 17. In general, these were the patients with the kind of history that most commonly precedes a laminectomy, with one or more attacks severe enough to require immobilisation during the few years preceding operation. The specimen illustrated in Figure 6 (*c*) was obtained from a patient aged thirty-eight, with a six months' history of sciatic pain immediately before operation, and a similar attack treated by a plaster jacket two years before. Figure 6, which in sum illustrates the extent of the variation in the imbibition characteristics of all the discs examined, is completed at (*a*) by the most hydrophilic of the normal series, obtained post-mortem from a subject aged sixteen.

DISCUSSION

A uniform finding throughout this series has been the lowered affinity for fluid shown by material from protruded or extruded discs irrespective of the patient's age. There seems little doubt that these changes must have preceded the mechanical derangement by a considerable period because no operation specimen, however short the clinical history, approached a normal degree of hydration at the time of its removal. It has already been stated that all operations were carried out when the patient was clinically in an acute phase, and there can be no question of the hydrophilia of the discs being substantially different at the time of actual protrusion. A further point is that no difference could be found between merely bulging discs and those

with a sequestrum lying free in the canal; in these latter cases, moreover, the sequestrum and the subsequent curettings from the disc space were in many instances examined separately and always gave closely comparable figures.

Two points, therefore, fall to be discussed, and will be taken in succession. First, the apparent conflict between the present findings and the commonly accepted view that disc protrusion is caused or contributed to by " hyper-hydration " or " engorgement " of the disc; and second, the effect which the described changes must have on the behaviour of the disc as a whole, before its derangement occurs.

The imbibition characteristics of discs, whether normal or abnormal, have usually been described in terms of the percentage increase in their weights occurring after their immersion in fluid. This increase is usually quite striking, as it was also in the present series of operation specimens. The discs shown in Figure 6 at (*b*) and (*c*), or the " average " example shown in Figure 4 (*a*), show increases ranging from about 40 per cent to over 100 per cent, and these figures are in accord with most published work. Comparison, on this basis only, with the relatively modest percentage increase of the normal disc (Fig. 4 (*b*)) could well lead to the conclusion that the deranged disc had in some way become more hydrophilic, and might be supposed to have generated some pressure in the intervertebral space, causing its protrusion. It is only when the true state of hydration is known by reference to the dry weight of material present that the fallacy of the above argument becomes apparent. When this is done it is clear that the high percentage increase of the abnormal disc indicates, not supersaturation, but an undue susceptibility to changes in environment; the large amount of fluid that it absorbs on removal from the body results from its inability to retain a normal amount when in the body. Far from " generating " an unusually high pressure, it reacts to increased pressure by losing an abnormally large volume of fluid.

The effects which this alteration in imbibition will have on the behaviour of the disc as a whole can be deduced with accuracy. Stresses transmitted through the vertebral bodies can be regarded as being resolved into two components: 1) a force balanced by the imbibition pressure of the nucleus, which will be wholly contributed by the nucleus; and 2) a residue which must be transmitted through or provided by the annulus. Hirsch (1951) pointed out that, if the nucleus " loses part of its faculty for producing an even distribution of pressure, the annulus is no longer capable of meeting even physiological demands made on it." It may be important also that in the normal disc at rest the " intrinsic pressure " of the nucleus (Petter 1933) is greater than the forces to be transmitted, and the annulus of such a disc would therefore be in tension and would experience compression only during activity. A reduction in the imbibition pressure of the nucleus will have three effects: 1) a greater proportion of the total strain will be thrown on the annulus; 2) the character of the strain may change from alternating tension and compression to unrelieved compression; and 3) under conditions of prolonged relaxation the disc will imbibe fluid, only to be unable to retain it when stress is reapplied. The first two factors are likely to cause gradual attrition of the annulus. The third factor will, on the application of strain, cause a sudden release of a volume of tissue fluid and a rapid redistribution of hydrostatic pressure, which will not easily be dissipated through the attenuated vascular communications of the disc.

I believe that these three factors provide in themselves a satisfactory explanation for most of the phenomena associated with disc protrusion. In particular, the sudden liberation of tissue fluid on loading a nucleus with a low imbibition pressure accords very well with the common history of the sudden onset of symptoms on first getting up in the morning, or with some trivial strain after a period of rest. There seems every reason to believe that repeated episodes of this kind would be sufficient to rupture an annulus which had already suffered damage from the action of the first two factors described above. Protrusion of the nucleus would follow, either gradually from weight bearing and movement or suddenly from another " hydrodynamic " episode. A minor corollary is that the reverse hydrostatic effect must

obtain when a pathological disc is freed from pressure, and may be the source of the "vacuum" disc phenomenon reported by Samuel (1948) and others.

CONCLUSIONS

The successive changes in imbibition pressure, which have been described above and illustrated in Figure 6, are exactly those which occur in an inorganic gel when it exhibits the phenomenon of "ageing." After maintaining their normal imbibition pressures for some time such gels pass first into a stage in which they can retain a near normal amount of fluid when unstressed, but cannot do so against pressure. A gradual decrease in the affinity between the phases then follows until finally the solid or disperse phase is able to bind to itself only a much reduced amount of its dispersions medium. The deranged discs in this series, therefore, exhibited changes which are to be ascribed to the loss of, or deterioration in, the muco-protein

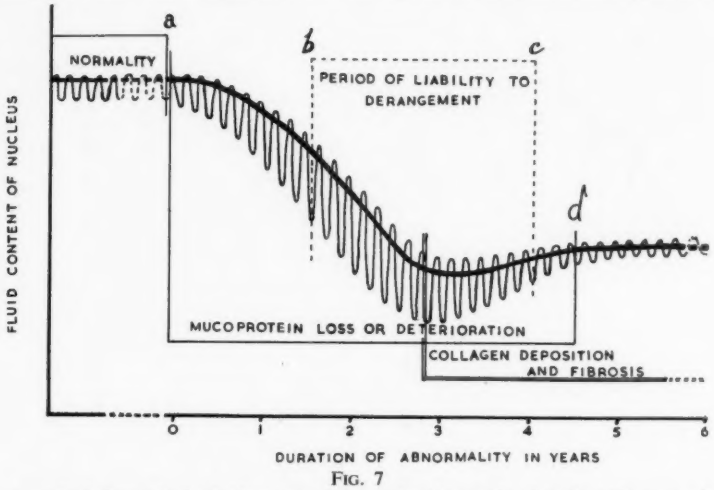

FIG. 7

A gradual change in the fluid content of the nucleus is postulated as the main cause of mechanical derangement. Over a period of some years the disc passes from its normal highly hydrated condition through a prodromal phase into a state in which protrusion or extrusion is likely. Thereafter, increasing deposition of collagen and fibrous tissue leads to clinical healing. Heavy continued line=effective imbibition index; wavy line=expected variation in the fluid content of the nucleus under normal loadings.

gel of the nucleus—that is, the same changes as have been shown by Sylvén (*loc. cit.*) and others to occur normally only with advancing years. If it be accepted, as I think it must, that these abnormalities preceded the mechanical derangement, then the etiology of disc displacement must include as a prime cause some unknown nutritional or biochemical factor which produces premature loss of, or deterioration in, the protein/polysaccharide complex of the nucleus. This is in agreement with the view put forward by Friberg (1948, 1950) after detailed anatomical and pathological work, that disc prolapse is to be regarded as evidence of earlier than normal degeneration.

The correlation of imbibition pressures with clinical history as summarised in Figure 6 makes it probable that the natural history of this premature ageing extends over a period of ten years or more. The progressive alteration in behaviour during that time can only be assumed, but it can be forecast with reasonable certainty from the data given in Figure 6, and expressed in terms of the effective imbibition index and of the expected variation in the fluid content of the nucleus under normal loadings (Fig. 7). It seems likely that the effective index represents very nearly the maximum degree of hydration which the nucleus can sustain within the intervertebral space; changes in water content under load would largely take place

below this level but should be roughly proportional to the imbibition differential. Having regard to the shape of the imbibition curves shown in Figure 5, I have assumed that the normal variation under load amounts to half the imbibition differential, and on this basis have represented in Figure 7 my conception of the natural history of a disc as it passes, in the course of several years, from normal, through a prodromal phase, to an " active " phase and then into clinical healing. This may serve to summarise the theory of disc protrusion which is here presented. At the left of the figure the disc's characteristics are normal, with a high water content, a high degree of elasticity and a relatively small volume change under load. Abnormal muco-protein loss or deterioration then sets in (*a*) and the nucleus becomes both less well hydrated and more susceptible to the effects of pressure. This is the prodromal phase which, on the time basis suggested by Figure 6, appears to last perhaps for eighteen months. At (*b*) the annulus has already suffered a period of attrition, hydrostatic effects are at their greatest, with a markedly lowered index and maximal volume changes in the nucleus under pressure; the disc enters the phase of liability to protrusion, and continues so liable until at (*c*) the muco-protein loss and the collagen replacement are such that major hydrostatic effects cease. At (*d*) the disc is still markedly dehydrated but, in virtue of the replacement of gel by fibrous tissue, is now less susceptible to pressure than a normal disc; no actual functional reconstitution of the nucleus has occurred, but clinically there is healing. Whether or not a mechanical derangement actually occurs during the phase (*b*) to (*c*) will presumably depend on the age and activity of the patient at this point, on the extent and speed of collagen and fibrous tissue deposition in the nucleus, on the relation of annular damage and fibrosis to hydrostatic effects, and perhaps on unknown biochemical factors. It may well be that the difference between normal ageing and the process described here may reside simply in the two phases of muco-protein loss and collagen deposition becoming out of step. One can well imagine that a slow, orderly muco-protein loss, with collagen replacement proceeding *pari passu*, would allow progressive " physiological " dehydration to occur without damage to the annulus and without hydrostatic effects appearing.

I have concerned myself here purely with the mechanical effects that can be explained by, or deduced as following, premature muco-protein loss in the nucleus. I have little information as to whether or not additional biochemical factors enter into the production of the actual derangement. It is possible, but I think unlikely, that the nucleus, having become deficient in gel, becomes susceptible to electrolyte or other changes in the blood. I have immersed different portions of the same deranged disc in normal plasma and in the plasma from a patient suffering an extremely acute derangement, without any difference appearing between the two. Another possibility is that, as the imbibition pressure of the nucleus decreases, osmotic pressure becomes more important. This must certainly be true to some extent, but I should expect that, by the time degeneration had proceeded thus far, tears in the annulus or in the cartilaginous end-plates would have rendered the disc ineffective as an osmotic system. It seems, moreover, unnecessary on general principles to postulate two abnormalities when one provides an adequate explanation of the known facts.

SUMMARY

1. Eighteen normal discs from cadavers and seventy-five specimens of abnormal disc material obtained at laminectomy have been compared.

2. Imbibition pressure, and not osmotic pressure, is the important factor in maintaining hydration of the nucleus, and the comparison has been based on this finding. The terms " free imbibition index " and " effective imbibition index " are suggested as being readily determinable means of expressing the functional efficiency of a nucleus.

3. A reduction in imbibition pressure was a constant feature of the specimens obtained at operation. No evidence was found to support the theory that hyper-hydration or engorgement of the nucleus plays a part in disc protrusion. A reduction in imbibition pressure can, however,

be expected in itself, by a combination of mechanical and hydrostatic effects, to cause disc derangements.

4. The reduced imbibition pressure of abnormal discs is related to abnormal loss of, or deterioration in, the protein/polysaccharide of the nucleus. The premature onset of, or some disturbance in, the normal ageing process is a prime cause of mechanical derangement of the disc.

I owe special thanks to Mr H. D. Griffith, B.A., F.R.S.E., Lecturer in Natural Philosophy, Aberdeen University, for bringing to my notice the facts concerning imbibition phenomena and for help in elaborating the theory of disc protrusion which is presented. My thanks are also due to Dr A. Lyall, Head of the Department of Clinical Chemistry, Aberdeen University, for the use of apparatus; to Mr A. M. Rennie for some of the operation specimens; to Professor J. Young for access to post-mortem material; and to Mr Jack Watt for the provision of the post-mortem specimens. The preparation of the figures was greatly assisted by Mr Topp of the Department of Clinical Photography.

Part of the work here described formed the basis of a short paper read before the British Orthopaedic Association in October 1954.

REFERENCES

BIRKETT, A. N. (1950): Injuries and Derangements of the Spinal Column. *In* Modern Trends in Orthopaedics, p. 299. Edited by Sir Harry Platt. London: Butterworth & Co. (Publishers) Ltd.

BUSH, H. D., HORTON, W. G., SMARE, D. L., and NAYLOR, A. (1956): Fluid Content of the Nucleus Pulposus as a Factor in the Disk Syndrome. British Medical Journal, **ii,** 81.

CHARNLEY, J. (1952): The Imbibition of Fluid as a Cause of Herniation of the Nucleus Pulposus. Lancet, **i,** 124.

COVENTRY, M. B., GHORMLEY, R. K., and KERNOHAN, J. W. (1945): The Intervertebral Disc: its Microscopic Anatomy and Pathology. Journal of Bone and Joint Surgery, **27,** 108.

DEUCHER, W. G., and LOVE, J. G. (1939): Pathologic Aspects of Posterior Protrusions of the Intervertebral Disks. Archives of Pathology, **27,** 201.

ECKERT, C., and DECKER, A. (1947): Pathological Studies of Intervertebral Discs. Journal of Bone and Joint Surgery, **29,** 447.

FRIBERG, S. (1948): Anatomical Studies on Lumbar Disc Degeneration. Acta Orthopaedica Scandinavica, **17,** 224.

FRIBERG, S., and HIRSCH, C. (1950): Anatomical and Clinical Studies on Lumbar Disc Degeneration. Acta Orthopaedica Scandinavica, **19,** 222.

GORTNER, R. A. (1938): Outlines of Biochemistry. Second edition. London: Chapman and Hall.

HAAS, S. L. (1939): Growth in Length of the Vertebrae. Archives of Surgery, **38,** 245.

HIRSCH, C. (1951): Studies on the Mechanism of Low Back Pain. Acta Orthopaedica Scandinavica, **20,** 261.

INMAN, V. T., and SAUNDERS, J. B. de C. M. (1947): Anatomicophysiological Aspects of Injuries to the Intervertebral Disc. Journal of Bone and Joint Surgery, **29,** 461.

KEYES, D. C., and COMPERE, E. L. (1932): The Normal and Pathological Physiology of the Nucleus Pulposus of the Intervertebral Disc. Journal of Bone and Joint Surgery, **14,** 897.

LINDAHL, O. (1948): Über den Wassergehalt des Knorpels. Acta Orthopaedica Scandinavica, **17,** 134.

MCBAIN, J. W. (1950): Colloid Science. Boston: D. C. Heath & Co.

MILWARD, F. J., and GROUT, J. L. A. (1936): Changes in the Intervertebral Discs following Lumbar Puncture. Lancet, **ii,** 183.

NAYLOR, A., and SMARE, D. L. (1953): Fluid Content of the Nucleus Pulposus as a Factor in the Disk Syndrome. British Medical Journal, **ii,** 975.

NAYLOR, A., HAPPEY, F., and MACRAE, T. (1954): The Collagenous Changes in the Intervertebral Disk with Age. British Medical Journal, **ii,** 570.

PEASE, C. N. (1935): Injuries to the Vertebrae and Intervertebral Disks Following Lumbar Puncture. American Journal of Diseases of Children, **49,** 849.

PETTER, C. K. (1933): Methods of Measuring the Pressure of the Intervertebral Disc. Journal of Bone and Joint Surgery, **15,** 365.

PUKY, P. von (1937): Über die Rolle der Zwischenwirbelscheiben bei Wirbelsäulenverkrümmungen. Archiv für klinische Chirurgie, **188,** 171.

PÜSCHEL, J. (1930): Der Wassergehalt normaler und degenerierter Zwischenwirbelscheiben. Beiträge zur pathologischen Anatomie und zur allgemeinen Pathologie, **84,** 123.

SAMUEL, E. (1948): Vacuum Intervertebral Discs. British Journal of Radiology, **21,** 337.

SAUNDERS, J. B. de C. M., and INMAN, J. T. (1940): Pathology of the Intervertebral Disk. Archives of Surgery, **40,** 389.

JUNGHANNS, H. (1931): Die Zwischenwirbelscheiben im Röntgenbild (Ihre Umbildungen während des Wachstums und ihre krankhaften Veränderungen). Fortschritte auf dem Gebiete der Röntgenstrahlen, **43,** 275.

SYLVÉN, B. (1951): On the Biology of the Nucleus Pulposus. Acta Orthopaedica Scandinavica, **20,** 275.

UEBERMUTH, H. (1929): Die Bedeutung der Altersveränderungen der menschlichen Bandscheiben für die Pathologie der Wirbelsäule. Archiv für klinische Chirurgie, **156,** 567.

IN MEMORIAM

STERLING BUNNELL
1882-1957

Dr Sterling Bunnell died on August 20, 1957, at his home in San Francisco.

Medicine has lost one of her most distinguished sons and many of us have lost a beloved friend. For Bunnell there were no national boundaries, and his friends and pupils—indeed all his pupils were his friends—came from all corners of the earth. We in the British Commonwealth mourn and salute him no less than do his fellow countrymen. Bunnell became a legend in his own day and this he achieved without consciously seeking fame. It came by virtue of his personal qualities, unhelped by association with a famous clinic or teaching school; during the greater part of his life Bunnell was not on the staff of a teaching hospital. What were the special gifts which led him to the high places of achievement and won for him the respect and affection of so many?

I believe that integrity, boundless enthusiasm and a deep humanity were his outstanding qualities. From his early days Bunnell was enthralled by the evolution of life, and his knowledge of biology was widely acclaimed. The introductory chapter of his book was written from his own knowledge of comparative anatomy. His many original contributions to the operative treatment of hand disabilities arose from his knowledge of the basic principles of the anatomy and function of the hand. It was this rather than his technical brilliance—although this he possessed in no small measure—that brought such rewarding results. He built upon rock and he could always give a clear and reasoned explanation for his opinions, revealing a depth of knowledge and wisdom that was at times humbling to his questioner.

Bunnell's enthusiasm was matched by his powers of endurance. His appointment during the war as consultant in hand surgery to the Surgeon General, United States Army, came when he was sixty years of age. He was responsible for the establishment of nine centres for hand surgery in Army hospitals, in which some 20,000 patients underwent reconstruction of the hand. For three years he was engaged in this exacting work. Patrick Clarkson has told me how he has seen Bunnell arrive at one of these centres, spend a full afternoon examining patients, sometimes devoting forty minutes to one difficult case, followed by an evening of seminars. Next morning there would be one or more complex repair operations before he left by plane at lunch time for his next assignment.

Immediately before beginning his Army work Bunnell, after much persuasion by Surgeon General Kirk, Messrs J. B. Lippincott and Mrs Bunnell, took a few months away from his practice to prepare his work for publication. As General Kirk said: " You will not always be with us and my young men must possess your knowledge."

Surgery of the Hand was an immediate and outstanding success, and established for all time this new and major school of surgery. The first edition, published in 1944, contained 734 pages; the third and last edition appeared in 1956 and had grown to 1,079 pages. New sections on ischaemic contracture in the hand and on the treatment of the rheumatic hand revealed Bunnell's active and searching mind even after he had passed his seventieth year. This book has probably had more influence and done more good than any other single work on surgery in the last fifty years.

Bunnell's love of the wild led him into many adventures. His film of the brown bears of Alaska, taken upon Admiralty Island, is a record not only of the bears but of Bunnell's complete fearlessness. He has told me how he and his son would photograph the bears—often with their cubs—at close range and the bears would turn about and peer for the source of the camera noise. When I asked if he carried a gun he replied: " No, I was photographing,

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not shooting, bears!" The story is told that when invited to lecture in Brazil in 1953 he accepted on condition that he might be given the opportunity to hunt the jaguar in the Matto Grosso. Visitors to his gracious home at 2000 Broadway will recollect the two alligators which roamed free in the garden.

Bunnell will be remembered by the many whom he met during his visits to Great Britain in 1946 and 1952. In 1946 he lectured at Guy's Hospital Medical School and at the Royal Society of Medicine. On both occasions the audience was so vast that there was no standing room. At Guy's the students performed prodigious feats of balancing in their efforts to find a single foothold, and at the Royal Society of Medicine the lecture was interrupted while the assembled company moved into a larger hall. Bunnell was deeply touched by these demonstrations. In 1952 Bunnell took part in the Joint Meeting of the Orthopaedic Associations of the English-Speaking World held in London and gave a notable address upon his work on ischaemic contracture of the hand. He had a deep affection for Great Britain, and his subsequent letters revealed how much these visits and the friendships he had made meant to him.

Bunnell was a man as complete in his affections as in his talents. "Fine art," said Ruskin, "is that in which the hand, the head and the heart of man go together." It was this rare integrity of person and profession, of skill and sympathy which enables us honestly to describe this distinguished American as a great man. R. G. P.

PROCEEDINGS AND REPORTS OF UNIVERSITIES COLLEGES, COUNCILS AND ASSOCIATIONS

GREAT BRITAIN AND IRELAND

NEW YEAR HONOUR

Sir Harry Platt—That the conferment of a baronetcy on Sir Harry Platt was only to be expected does not at all diminish the satisfaction felt by orthopaedic surgeons everywhere. In 1948 we had the pleasure of announcing his award of a knighthood, and in 1954 his election as President of the Royal College of Surgeons of England; the first orthopaedic surgeon to hold this high office, he fulfilled it with great distinction at a time of much difficulty. In the same year he concluded six years' tenure of office as President of the International Society of Orthopaedics and Traumatology, achieving both consolidation and development. The brilliant career, the many achievements, and above all the remarkable judgment and wisdom, are so well known and have been so often recorded in the pages of our Journal that we will content ourselves with merely congratulatory messages—first to Sir Harry on this recognition of his worth and secondly to ourselves on the distinction bestowed on one of our most respected brethren.

UNIVERSITY OF EDINBURGH

APPOINTMENT OF PROFESSOR OF ORTHOPAEDIC SURGERY

We are pleased to be able to report that Mr J. I. P. James, Assistant Director of Studies in the Institute of Orthopaedics of the University of London, and Consultant Orthopaedic Surgeon to the Royal National Orthopaedic Hospital, has been appointed to the Chair of Orthopaedic Surgery in the University of Edinburgh, on the lamented retirement of Sir Walter Mercer. The appointment will give great satisfaction to Mr James's many friends here and overseas.

Elliot & Fry

MR J. I. P. JAMES

Mr James received his medical education at University College, London, and its attached hospital, at each of which he gained scholarships. He served in all the general surgical posts up to that of surgical registrar in his own teaching hospital. This, alone among the twelve London undergraduate teaching hospitals, lacks an orthopaedic department, and it was as house surgeon and later registrar at the Royal National Orthopaedic Hospital that he became attracted to the speciality for whose discipline an exceptionally good general training had prepared him.

Equipped with the London M.S. and English F.R.C.S., he joined the Army in the second world war as a surgical specialist and was soon parachuted into Jugoslavia as a forward surgeon with guerilla forces. Surprisingly he survived both friend and foe to become surgeon with the Parachute Brigade in Greece and, later, to command a surgical division in the rank of Lieutenant-Colonel.

After the war, with four others, he was elected to the consultant staff of the Royal National Orthopaedic Hospital; and he took a leading part in the development of the Institute of Orthopaedics, which raised that hospital to the stature and the status of a postgraduate teaching hospital. Soon after his appointment he secured a Rockefeller Fellowship for a year's study in the United States of America, where he visited many centres, making a particular study of hand surgery and of scoliosis. Special clinics in these subjects were established for him at the Royal National Orthopaedic Hospital on his return, and the fruit that they have borne is well known. In 1956 he was appointed Consultant in Orthopaedics (Hand Surgery) to the Royal Navy.

Mr James has been active in the affairs of the British Orthopaedic Association, of which he has been Honorary Secretary since 1955.

The Professor's future duties include: undergraduate teaching; the conduct, supervision and development of research; examining; the charge of not less than one hundred beds in the Princess Margaret Rose Orthopaedic Hospital and the Royal Infirmary; the duties of Adviser in Orthopaedics to Scottish South-Eastern Regional Hospital Board; and membership of various University governing and advisory bodies. The post is reputedly part-time, but Mr James would do well to sell his boat.

We believe that Mr James will find much goodwill and a kindly welcome; and we suspect that this Sassenach (or Saxon), precipitated into the Athens of the North, will emerge as unscathed as he did from his Jugoslavian adventure and from the Grecian capital itself. All orthopaedic surgeons will wish him good fortune. There is no doubt that he has something to give Edinburgh, and Edinburgh has something to give him.

UNIVERSITY OF LIVERPOOL

LADY JONES MEMORIAL LECTURE

On Tuesday, October 22, 1957, in the New Medical School of the University of Liverpool, Professor José Valls of the University of Buenos Aires delivered the thirteenth Lady Jones Memorial Lecture. His subject was "The Charcot Knee."

Having dealt with the history, the etiology, and more particularly with the histopathology, Professor Valls proceeded to deal with treatment. He had operated on ten knees, and it was in doing this that he had acquired the material for the microscopic examinations. He used the compression technique of Key, developed by Charnley, and in six cases bony fusion had been achieved. In the others fibrous union occurred, and since good alignment was maintained the patients were able to walk satisfactorily wearing a short knee splint.

DEGREE OF M.Ch.Orth.

Eight candidates were successful in gaining this degree. They were: F. H. Beddow, Liverpool, England; L. Bryer, Johannesburg, South Africa; F. A. Ducharme, Ottawa, Canada; T. H. F. Gillespie, Nairobi, Kenya; A. M. B. Ismail, Kuala Lumpur, Malaya; G. A. Jose, Adelaide, Australia; H. K. S. Murthy, Mysore, India; and H. S. Sabbak, Baghdad, Iraq.

It was not quite possible to recommend any of the candidates for a gold medal, but the examiners were so impressed with the clinical work of Mr F. H. Beddow that they spontaneously contributed to what was named for the first time the "Examiners' Prize."

The External Examiners were Mr Norman Capener of Exeter and Mr H. Osmond-Clarke of London. The Internal Examiners were Professor Bryan McFarland, Mr Norman Roberts and Mr Eric Wardle.

NORTH-EAST METROPOLITAN REGIONAL ORTHOPAEDIC CLUB

A meeting was held at Southend General Hospital on October 26, 1957, under the joint chairmanship of Mr J. H. Shelswell and Mr D. Ll. Evans. The meeting was devoted to a review of cases.

Fracture of lateral femoral condyle—A fracture of the articular surface of the lateral condyle of the femur had been treated by open reduction and fixation with two Pidcock's pins, the pins being removed five months later. An excellent functional result had been obtained.

Infantile kyphosis—Thoracic kyphosis occurred in an infant of five months, who was said to have been paralysed temporarily in both legs soon after birth. The deformity was first noticed three weeks later. Radiographs showed absence of three thoracic vertebral bodies with some evidence of anterior calcification. At five months there was no paralysis and the child appeared healthy. The diagnosis was thought to rest between a congenital deformity or a neo-natal osteomyelitis, but the radiographic appearances could not be fully explained on this basis.

Elbow injuries—In a case of compound fracture involving the left elbow there was persistent anterior displacement of the radial head. During a lively discussion the methods of treatment suggested included a formal arthroplasty, excision of the radial head alone, and excision of the radial head combined with intramedullary fixation of the ulna.

In another case of compound comminuted fracture involving the right elbow treatment by excision of the head of the radius and intramedullary fixation of the ulna had been carried out. This was followed by cross union of the radius and ulna, but non-union of the ulnar fracture, which required a bone graft a year later. The patient had regained a stable painless elbow but almost no rotation. It was agreed that further treatment would not be justifiable.

Pathological fracture—A pathological fracture of the tibia, from a football injury, in an eleven years old boy was shown. The fracture had failed to unite, and the fragments had angulated forwards after six months' immobilisation. A bone graft applied nine months after the injury fractured three months later. Histology was indefinite but suggested a localised fibrous dysplasia of bone, although congenital pseudarthrosis of the tibia was also suggested.

Case for diagnosis—A Merchant Navy officer complained of chronic swelling and tenderness of the lower part of the shin after a kick at football. All investigations were negative and a biopsy revealed no definite pathology. No other bone was involved. The case was regarded as one of chronic low-grade osteomyelitis, possibly due to a tropical infection.

Joint transplant—In a case of circular-saw injury of the hand a transplant of the entire metacarpo-phalangeal joint from the little finger had been carried out, seemingly with success. The joint was still functioning fifteen months later, although it was noted that avascular necrosis of the metacarpal head had occurred, so far without deformity. It was suggested that if deformity occurred a cancellous graft should be placed under the articular surface.

NORTH-WEST METROPOLITAN ORTHOPAEDIC CLUB

A meeting of the North-West Metropolitan Orthopaedic Club was held at the Royal Air Force Rehabilitation Centre, Headley Court, on November 9, 1957, by the kind invitation of Air Commodore L. M. Crooks.

Tendon injuries—*Air Commodore G. H. Morley* stressed the importance of getting the injured finger into a good position before operation. Lateral incisions were used, the pulley slings being made from the tendon sheath opposite the middle of the phalanx. Correct tension was obtained by attaching the lower end first and then the upper. Tension on the tendon was relaxed by flexing the wrist, not the finger. The importance of post-operative elevation was emphasised. Of forty flexor profundus grafts for division of the flexor sublimis and profundus, excellent or good results had been obtained in twenty-four, and fourteen were moderate. Two fingers required amputation. For severance of the flexor pollicis longus a graft from wrist to tip of thumb was advocated unless the site of division was near the tip. Twelve out of sixteen patients had good results.

Electrodiagnosis—*Squadron Leader C. B. Wynn Parry* recalled that by electrical tests it was possible to detect at an early stage 1) nerve degeneration, 2) innervation, 3) nerve anomalies (fairly common in the hand) and 4) the exact site of a lower motor neurone lesion. Faradism could cause contraction in denervated muscles; so faradic or galvanic tests were unreliable. Strength-duration curves were more reliable. By these curves delayed progress could be detected early (neuroma), and early exploration was then to be encouraged. Electromyography using surface suction electrodes or needles was described. In cases of nerve injury discrete activity could be demonstrated, and recovery could be detected by this means up to three months before clinical detection was possible. A recovering muscle might act first as a synergist but not as a prime mover. Fibrillary rhythmic activity of degenerated muscle was demonstrated.

Clinical cases—The following cases were shown: 1) A child with an infective lesion of the right lateral femoral condyle. Premature fusion of the outer femoral epiphysis had taken place. The problem of further treatment was raised. 2) A case of staphylococcal osteitis of the first and second lumbar vertebrae with two sinuses from a psoas abscess. 3) A case of staphylococcal infection of the cervical spine, skull and sterno-clavicular joint. 4) Osteoid osteoma near the elbow. 5) Two cases of posterior subluxation of the shoulder, without history of injury; one patient was symptomless.

SOUTH-EAST METROPOLITAN REGIONAL ORTHOPAEDIC CLUB

A meeting was held at the Royal Air Force Rehabilitation Unit, Headley Court, on October 26, 1957. The commanding officer, Wing Commander J. A. MacCarthy, welcomed the visitors, who were later shown round the Unit where about forty patients were undergoing treatment. The patients' disabilities ranged from fractured femur and ankylosing spondylitis to nerve injuries in the hand and poliomyelitis. Particular admiration was expressed by members for the hydrotherapy and occupational therapy departments, and it was clear to all that the general spirit of the Unit was excellent.

Rehabilitation after injury—*Squadron Leader P. J. R. Nichols* outlined the principles of rehabilitation and the organisation of the Unit, and illustrated these principles by describing the treatment of lower limb fractures. Since the end of the war about 30,000 patients had been treated at the three medical

rehabilitation units, and of that number 70 per cent had returned to full duty and 25 per cent to modified duty; 5 per cent had been invalided. Patients were allocated to remedial classes appropriate to their condition, and the programme consisted of a five-hour working day divided between class exercises, physiotherapy, hydrotherapy, occupational therapy, organised games, walks, etc. Stress was laid on the necessity of obtaining the patients' full active co-operation at an early stage in treatment, and individual attention was given during classes as well as at the weekly review clinic. Under these conditions very few patients had been found " unsuitable " for treatment in the Unit.

When a patient is ultimately invalided it devolves upon the Unit to establish the chain of resettlement, and no patient in this category was discharged without final arrangements for his continued treatment, training or employment.

Referring to lower limb fractures Squadron Leader Nichols said that the chief problems were mobilisation of the ankle in fractures of the tibia, quadriceps development in knee injuries, and mobilisation of the knee in fractures of the femur. Since in all cases walking was affected, the object of treatment from the outset was to achieve good power, good movements, good co-ordination, and the elimination of unequal stresses and strains from improper balance. It was essential to treat the patient as an individual and not as a given type of fracture, because the conflicting incentives to return to duty varied in every case.

In an analysis of 400 fractures of the tibial shaft, of which 150 were subjected to open operation at some stage, it was found that the total disability time in simple fractures was twenty-five weeks, in compound fractures thirty weeks, and in operation cases thirty-five weeks. The mobilisation time—the interval between clinical union and final return to duty—was prolonged by any factor that delayed union, and the earlier a patient could be transferred to the Unit before clinical union, the shorter was the mobilisation time. The problems of stiffness of the ankle, oedema, and persistent limp were directly related to lack of regular exercises and proper balanced walking in the stage of immobilisation.

Hand injuries—*Squadron Leader C. B. Wynn Parry* demonstrated a number of cases of hand injury, including examples of ulnar nerve division, crush injury and the " spring double " (division of both nerves and all tendons at the front of the wrist).

General discussion—In the general discussion *Mr H. L.-C. Wood* enquired about the treatment of Sudeck's osteodystrophy and was told: " Three aspirins, a wax bath and hard work "—a suggestion upon which no one present could improve. The diversity of opinion on aspiration of the knee was remarkable: some surgeons never aspirated at all, whereas some aspirated on the slightest provocation; the physicians compromised by aspirating the haemarthroses and dispersing the simple effusions by pressure, immobilisation and exercises. The officers of the Unit had had no experience of early x-ray therapy for crush injuries of the hand, but *Squadron Leader Wynn Parry* stressed the advantages of splinting in the position achieved at the end of each treatment.

IRISH ORTHOPAEDIC CLUB

The autumn meeting of the Irish Orthopaedic Club was held in Dublin on November 16, 1957, at Dr Steevens' Hospital and the Temple Street Children's Hospital.

Anterior spinal fusion—*Mr W. H. deWytt* discussed anterior spinal fusion, with special reference to the difficulties he had encountered in forty-five cases. In his series he had found it necessary to place a large tibial graft well forward, particularly when there was acute kyphosis. He thought it necessary to mobilise the aorta from the second to the tenth intercostal vessels, but had had difficulty with the leash of vessels extending from the aorta to the apex of the kyphos. Mr deWytt believed that the anterior approach through the chest was the procedure of choice in the operative treatment of paraplegia. Finally he described his method of anterior exposure and fusion in the lumbo-sacral region. In discussion most members agreed that the anterior fusion, with or without decompression, was a very real advance in treatment, but they thought that lateral rhacotomy still had an important place in the relief of Pott's paraplegia, and they did not all agree with Mr deWytt about anterior fusion in the lumbar region.

Approach to the sacro-iliac joint—*Mr M. G. A. Little* showed a film on the approach to the sacro-iliac joint by splitting off the outer table of the posterior part of the ilium, the gluteal muscles being undisturbed on its outer surface.

Clinical cases—Clinical demonstrations were given by *Mr Arthur Chance*, *Mr J. Boyd Dunlop*, *Mr C. McDowell* and *Mr P. Fitzgerald*; and in the afternoon by *Mr J. P. Shanley* (who showed the late results of treatment of septic arthritis in infancy and adolescence), *Mr P. MacAuley* and *Mr J. Gallagher*.

UNITED STATES OF AMERICA

JOINT MEETING OF THE ORTHOPAEDIC ASSOCIATIONS OF THE ENGLISH-SPEAKING WORLD

The joint meeting of the orthopaedic associations of the English-speaking world will be held in Washington, D.C., from May 11–16, 1958. The provisional programme is as follows.

Monday, May 12, 2–5 *p.m.* (*At the Shoreham Hotel*)

Orthopaedic Problems in Children

Infantile Scoliosis	J. I. P. James, London, England G. C. Lloyd-Roberts, London, England
Congenital Pseudarthrosis of the Tibia	H. B. Boyd, Memphis, Tennessee F. P. Sage, Memphis, Tennessee
Congenital Absence of the Fibula	C. A. Laurens, Toronto, Canada A. W. Farmer, Toronto, Canada
Paraplegia in Children	P. L. Norton, Boston, Massachusetts J. J. Foley, Boston, Massachusetts
Epiphysial Stapling	W. P. Blount, Milwaukee, Wisconsin
Congenital Muscular Torticollis	G. J. Garceau, Indianapolis, Indiana W. L. Franklin, Indianapolis, Indiana

Tuesday, May 13, 9 *a.m.*–1 *p.m.* (*At the National Institutes of Health, Bethesda, Maryland*)

Symposium on Arthritis

Factors which Predispose to Osteoarthritis	Leon Sokoloff, N.I.H., Washington, D.C.
Hemiarthroplasty of the Knee Using a Space-occupying Prosthesis	D. L. McIntosh, Toronto, Canada
Mould Arthroplasty of the Knee	O. E. Aufranc, Boston, Massachusetts W. A. Jones, Boston, Massachusetts
Rheumatoid Arthritis of the Hand	O. J. Vaughan-Jackson, London, England
Surgical Treatment of Rheumatoid Deformities of the Hand	D. C. Riordan, New Orleans, Louisiana S. B. Fowler, Nashville, Tennessee
Rehabilitation of the Hand in Rheumatoid Arthritis	J. G. Kuhns, Boston, Massachusetts
Spinal Osteotomy	W. A. Law, London, England
Hazards of Steroid Therapy	Joseph Bunim, N.I.H., Washington, D.C.

2–3 *p.m.* (*optional*)

Tour of National Institutes of Health

Tour of Tissue Bank, National Naval Medical Center, Bethesda, Maryland

Wednesday, May 14, 9 *a.m.*–12.30 *p.m.* (*At the Shoreham Hotel*)

Symposium on Fractures

Blood Supply of Tibia in Relation to Healing of Fractures	Ian MacNab, Toronto, Canada
Non-union of Fractures of Long Bones	E. F. Cave, Boston, Massachusetts
Delayed Reduction of Fractures	J. R. Moore, Philadelphia, Pennsylvania
Central Dislocation of the Hip	G. F. Pennel, Toronto, Canada
Cervical Spine Injuries: Entities and Indications for Fusion	J. N. Garber, Indianapolis, Indiana

The Luscka Joints of the Cervical Spine: Anatomy, Physiology and Pathology	E. L. Compere, Chicago, Illinois
The Stored Homograft: Experimental Date and Clinical Implications	G. W. Hyatt, National Naval Medical Center, Bethesda, Maryland

2.30–3.30 *p.m.* (*optional*)

Symposium: Surgical Implants in Modern Surgery

Co-Chairmen: Mr Norman Capener, Exeter, England
Dr Joseph S. Barr, Boston, Massachusetts

Members of Panel: To be announced

Thursday, May 15, 9 *a.m.*–1 *p.m.* (*At the Walter Reed Army Medical Center*)

Symposium on Bone Tumours

The Development of the Histological Classification of Malignant Bone Tumours	H. A. Sissons, London, England
Surgical Treatment of Bone Sarcomas	Col. M. S. Thompson, Walter Reed Army Medical Center Col. S. J. Ritchey, Walter Reed Army Medical Center
Osteogenic Sarcoma	K. C. Francis, New York R. L. Cunnett, New York
Chondrosarcoma	Roland Barnes, Glasgow, Scotland
Recent Experiences with Treatment of Osteogenic Sarcomas	W. T. Green, Boston, Massachusetts
Prosthetic Replacement After Local Resection of Bone Tumours	H. Jackson Burrows, London, England J. T. Scales, London, England
Orthopaedic Aspects of Myelomatosis	D. Lloyd Griffiths, Manchester, England
Parosteal Osteoma of Bone	M. M. Copeland, Washington, D.C. C. F. Geschickter, Washington, D.C.

2–3 *p.m.* (*optional*)

General Biological and Field Theory of Bone Tumours	L. C. Johnson, A.F.I.P., Walter Reed Army Medical Center
Tour of Armed Forces Institute of Pathology	
Tour of Walter Reed Army Hospital	

Friday, May 16, 9 *a.m.*–12.30 *p.m.* (*At the Shoreham Hotel*)

Lumbosacral Interbody Fusion	G. F. Dommisse, Pretoria, South Africa
Changes in the Hip Joint after McMurray's Osteotomy	G. V. Osborne, Lancaster, England
The Organisation of the Components of Connective Tissue Matrices that Normally Calcify	R. A. Robinson, Baltimore, Maryland
The Relationship Between Molecular Ultrastructure and the Property of Calcificability in Bone	Melvin Glimcher, Boston, Massachusetts
The Anatomy of Bone Cysts	Robert Kindred, Charlottesville, Virginia L. C. Johnson, A.F.I.P., Walter Reed Army Medical Center
Rheumatoid Synovial Cyst of the Hip	M. B. Coventry, Rochester, Minnesota H. F. Polley, Rochester, Minnesota
The Relationship Between the Types of Sickle Cell Disease and Bone Changes	J. S. R. Golding, Jamaica, West Indies

TOURS FOR MEMBERS ATTENDING THE JOINT MEETING

Tours have been arranged to certain orthopaedic centres in the United States and in Canada during the two weeks after the Washington meeting. These are as follows:—

Tour 1.

May 17 Visit to Johns Hopkins Hospital, Baltimore, Maryland, and the Alfred I. duPont Institute, Wilmington, Delaware.
May 18 Rounds at Children's Seashore House at Atlantic City.
May 19 Clinical Programme at University Hospital, Philadelphia.
May 20 Clinical Programme at Hospital for Special Surgery, New York City.
May 21 Clinical Programme at New York Orthopaedic Hospital.

Tour 2. (May include Tour 1)

May 22 Clinical Programme by orthopaedists of Toronto.
May 23 Clinical Programme by orthopaedists of Montreal.

Tour 3. (May include Tours 1 and 2)

May 24 Clinical Programme in Boston, Massachusetts.
May 25 Sightseeing in Boston, Massachusetts.
May 26 Clinical Programme in Boston, Massachusetts.
May 27 Clinical Programme in New York at Hospital for Joint Diseases.
May 28 Sail in *Queen Elizabeth* to England.

ITALY

ITALIAN ORTHOPAEDIC AND TRAUMATOLOGICAL ASSOCIATION

The forty-second congress of the Italian Orthopaedic and Traumatological Association was held in Rome on October 29–31, 1957, under the presidency of Professor C. Marino-Zuco.

The main subjects for discussion were: 1) traumatic epiphysial separations, and 2) metaphysial fractures in infancy and childhood. The principal speakers on the first subject were *Professor A. Rampoldi* and *Professor M. Boni* of the Rome Orthopaedic Clinic; and on the second subject *Professor E. De Marchi*, of the Orthopaedic Clinic of Bari.

INTERNATIONAL SOCIETIES

INTERNATIONAL SOCIETY OF ORTHOPAEDIC SURGERY AND TRAUMATOLOGY

The seventh congress of the International Society of Orthopaedic Surgery and Traumatology (S.I.C.O.T.) was held in Barcelona from September 16–21, 1957. The attendance was nearly one thousand, including 636 members. The beautiful city, Catalan hospitality and the kindly climate combined to make this a very happy occasion. Professor José Valls (Buenos Aires) presided.

The treatment of flail joints—*Professor G. Hohmann* (Munich) discussed the many causes, including congenital dysplasia, constitutional tissue defects, injuries, nervous diseases, arthritis or mechanical derangements, stressing that a flail limb is usually the consequence of combined muscular and mechanical forces rather than of hypermobility of a particular joint.

Dr Manuel Bastos Ansart (Barcelona), discussing the surgical treatment of flail foot, said that for foot deformities and paralysis of the knee it was necessary to fuse the whole posterior tarsus, and preferably the ankle as well after a modification of Lorthioir's method.

Professor O. Scaglietti and *Dr G. Fineschi* (Florence) recalled that flail limb, interpreted strictly, had rarely any cause in the upper limb except loss of bone at a joint. Joints that were flail only when inactive, and that became controlled with muscular action, usually needed little more than exercise and re-education, but inadequate muscle control called for surgical treatment; only arthrodesis could yield constant and permanent results. If there were only slight loss of bone simple arthrodesis by apposition of cancellous bone usually sufficed, but with considerable loss of bone reconstruction with grafts was necessary.

Dr J. Cabot Boix (Barcelona) said it was necessary to consider joints in which the three kinetic elements were affected: muscle, bone and joint. He considered stabilisation of the knee, briefly reviewing interventions on all the ligaments and emphasising the particular importance of a sound medial [medial collateral, internal lateral] ligament.

Dr Knud Jansen and *Dr Lis Zachariea* (Copenhagen) described methods of tendon transfer in cerebral palsy and the requirements for success.

Professor C. Marino-Zuco and *Dr A. Rampoldi* (Rome) described the treatment of flail joints caused by poliomyelitis. They practised arthrodesis even during growth and showed that, if the epiphysial cartilage were respected, growth proceeded normally in shoulder, knee and foot.

The influence of growth on the sequelae of bone and joint injuries—*Professor J. Trueta* (Oxford), discussing the physio-pathology of the epiphysial cartilage and referring to the studies of the vascular arrangement near the cartilage made by the Oxford school, explained that, whereas the vessels near the epiphysial side of the plate had a nutritional role, those of the metaphysial side had calcification as their only function; and thus their interruption caused thickening of the growth plate like that of rickets, while discontinuation of the epiphysial circulation killed the germinal cells and, consequently, the whole of the plate. Professor Trueta discussed also the response of the epiphysial cartilage to pressure, and showed that lack of pressure caused an increased metaphysial blood flow which was responsible for the early closure of the plate, whereas excessive pressure caused metaphysial ischaemia and thus thickening of the growth plate. He concluded that the behaviour of the epiphysial cartilage would, in the main, depend (except in cases of traumatic disorganisation) on the distribution of pressures through the plate and, in the last instance, on the characteristics of the adjacent blood flow.

Dr P. L. Chigot (France), who had observed 3,997 fractures of the limbs between 1950 and 1955, recalled that apparently severe injuries in children often left little or no disability in adult life. The correction of deformity with epiphysial growth was perhaps influenced by pressure, perhaps by circulatory changes. Dr Chigot considered the mechanical factors in detail. The long-recognised lengthening after fractures was often modest; it did not affect the outlook, and there was good functional adaptation. It was a common if not constant happening. The lengthening was probably often ephemeral, being corrected by precocious cessation of growth. It was greatest if the position of the fragments was imperfect. In 205 cases of femoral fracture it was found that shortening of two to thirty millimetres from imperfect reduction was almost always compensated fairly quickly, and that consolidation without shortening led to excessive length of five to ten millimetres in 50 per cent, appearing from the sixth month. It happened more often after the age of three years. It could be explained by Trueta's ideas.

Dr Walter P. Blount (Milwaukee) advocated prompt surgical treatment of linear and angular deformities from epiphysial injury by methods of growth stimulation or osteotomy on the short side, or by retardation of the long side, or by a combination. Epiphysiodesis was satisfactory if there were no danger of over-correction. Stapling was the method of choice if adequate equalisation were possible and should be carried out as soon as expedient after the age of eight years. Most important precautions were: 1) not to bury the staples beneath periosteum; 2) not to damage the epiphysial plate during their insertion or removal; 3) three-monthly examination—including observations of gait and of standing and sitting heights, and also accurate radiographic measurements when calculated growth of the lower extremities exceeded one centimetre; 4) prompt removal of staples when length had become equal (there would be brief acceleration at first but this would be partly or completely compensated for by premature closure); 5) correction of angular deformity by asymmetrical stapling; 6) to aim at over-correction if the cause of deformity would persist; 7) correction of angular deformities in shortened limbs in children by osteotomy, not by stapling.

Professor Bryan McFarland (Liverpool) said that deformity from injury in children might be caused not only by involvement of the epiphysial plate but also by scarring or by malunion of fractures with consequent alteration of the line of gravity through the epiphysis. These should be dealt with before deformity occurred. *Dr J. Böhler* (Linz) was understood to say that these complications were not seen in his Clinic. *Professor U. Camera* (Turin) treated traumatic arrest of growth at the lower femoral epiphysis by excising its fellow, which he thought more certain than arrest by grafting (Phemister) or stapling (Blount).

Professor C. Casuccio (Padua) had confirmed all Trueta's observations on the vascularisation of bone with growth. He referred to growth of the upper end of the femur after the fairly frequent obstetrical epiphysial separation, which rapidly underwent malunion; the deformity would slowly disappear, and surgical intervention was absolutely wrong. *Dr Jean Judet* (Paris) said that increased growth after fracture was inconstant and did not exceed two centimetres; greater shortening than this required surgical correction. Growth would correct lateral displacements, but not angular ones, which would ultimately lead to arthrosis. In general, nature rarely corrected negligence.

Dr A. Langenskiöld (Helsingfors) had found that a condition radiologically and histologically indistinguishable from osteochondritis dissecans in man could be produced by separating and then replacing a fragment of cartilage of the femoral condyle of a very young rabbit; and he thought that a cartilaginous injury in an infant, though causing few symptoms at the time, might, with growth, cause severe sequelae.

Professor C. Mau (Heidelberg) observed that, if there was much shortening from disturbance of growth of the lower extremity of the femur of the young child, troubles might be encountered at the upper end of the femur; coxa valga, anteversion, or an unusually forward or low position of the lesser trochanter. The etiology was being studied.

Professor C. Marino-Zuco, *Dr A. Rampoldi* and *Dr M. Boni* (Rome) gave statistics of 700 cases of epiphysial separation observed in the Orthopaedic Clinic of Rome and reviewed after one year to twenty-five years. Experimental investigations of the pathology and the process of consolidation were described, including histochemical and radioactive isotope studies. Except in special instances the prognosis was good and the treatment conservative. Operation was reserved for cases in which there was final deformity.

Dr R. S. Reich (Cleveland, Ohio) advocated Blount's epiphysial stapling in spite of occasional unsatisfactory results which he ascribed to injury at the time of removal. Epiphysiodesis by Phemister's method or by Warren White's modification might be preferable for symmetrical arrest. *Dr E. Sorrel* and *Mme Sorrel-Déjerine* (Paris), while agreeing that many childhood traumatic deformities are corrected by growth, emphasised the importance of recognising those that are not. From the records of 1,789 fractures of the upper limb in children, they concluded: first, that epiphysial separation or involvement of an epiphysial cartilage in a fracture hardly ever led to disturbed growth unless metal were introduced near the cartilage; secondly, that during growth bone had a remarkable tendency to regain its normal form, a broken bone tending to regain and then maintain its normal length. *Dr J. Verbrugge* (Ghent) said that growth could not be depended upon always to restore shape and length. Perforation of an epiphysial cartilage was dangerous, except at the lower end of the humerus, which he had often nailed with impunity.

Giant-cell tumours and their treatment—*Mr R. Barnes* (Glasgow) spoke of the importance of distinguishing the true giant-cell tumour from such conditions as simple bone cyst, aneurysmal bone cyst, non-osteogenic fibroma and benign chondroblastoma. He condemned x-ray treatment without biopsy, especially now that the spine and pelvis had become accessible to this. It was difficult to distinguish between indolent and active giant-cell tumours.

Dr H. Boyd (Memphis) agreed with the difficulty of distinguishing the true giant-cell tumour but relied on staining for acid phosphatase; the stromal cells of giant-cell tumours, alone among these conditions, revealed it. *Professor I. Miki* (Tokio) spoke of the bad prognosis in the elderly and the histological evidence of malignancy as suggested by the poor lining of the vessels, the type of giant cells, variations in size of the stromal cells and mitoses. *Professor U. Camera* (Turin) considered that, at first, giant-cell tumours were benign and needed conservative treatment. Later, they needed more drastic treatment with reconstruction by grafts. He showed replacement of an extensive growth by fragments from a large exostosis.

Mr N. Capener (Exeter) described five cases of large giant-cell tumours in weight-bearing bones satisfactory ten years after operation. Three involved the upper end of the tibia, one the lower end of the tibia, and the other the lower end of the femur. The first four were treated by curettage and packing, the fifth by excision.

Dr D. Dahlin (Rochester, Minnesota) had been unable to find any adequate published series of cases treated by x-ray methods. They should not be used without a special indication. There had been five cases of malignant transformation after this treatment. Of 109 cases at the Mayo Clinic, eleven had undergone malignant change, in eight of them after a latent interval of years; the new tumour was a fibrosarcoma in seven cases and an osteosarcoma in one. Among thirty-one patients treated by curettage, and among thirty-one patients treated by curettage and x-ray, the rate of recurrence was practically identical, namely about 50 per cent. He concluded that deep x-ray treatment was a bad primary treatment and did not prevent recurrence of a tumour that had been curetted.

Dr P. Rubiés Trias (Barcelona) considered that curettage and grafting gave better results than deep x-ray treatment. Recurrence in a limb called for amputation. *Dr J. Sanchez Lucas* (Barcelona) spoke of the histological characteristics of giant cells.

Professor O. Scaglietti (Florence) distinguished on clinical, radiological and pathological grounds three types, which he called benign, locally malignant and malignant, the last extremely rare. The indications of malignancy were a rapid clinical course and a transitional histology. In the first type curettage should be avoided because of the frequent recurrence; radiotherapy should also be avoided because it could not be relied upon to cure and might produce malignancy. Treatment of the locally malignant type was resection, with bone grafting if this were important for limb function. Very large tumours and those in the elderly should be treated by amputation.

Dr Philip Wilson (New York) had studied thirty-three cases. He favoured resection when this was possible without too much functional disturbance, for instance at the upper end of the fibula or in scapula, talus, phalanx or metatarsal. At major joints curettage should be tried once, with chemical cauterisation and insertion of chips from the bone bank. Though diagnosis must be based

on biopsy there had been diminishing confidence in the pathologist's ability to distinguish between benign and malignant. Six cases out of thirty-three had pursued a malignant course. He confirmed the danger of promoting malignant change by x-rays.

Professor R. Zanoli (Bologna) advocated curettage in cases of giant-cell tumour, resection for giant-cell sarcoma and reservation of amputation for recurrences only. He had used metallic prostheses with good results, especially at the upper end of the humerus, and also grafts, of which Putti's cortical graft was specially recommended.

Professor J. Valls (Buenos Aires) spoke of the unreliability of both histological appearances and clinical behaviour in the early diagnosis of malignancy. Recurrence was possible after any type of treatment, and more and more cases were coming to light. He quoted Thomson and Turner-Warwick's figures of 45 per cent recurrence after curettage and 49 per cent after irradiation. Professor Valls advocated resection and grafting whenever possible and reservation of x-ray treatment for tumours inaccessible to surgery.

The surgical treatment of osteoarthritis of the hip (arthroplasty excluded)—*Professor J. Trueta* (Oxford) opened with an account of the main pathological features of osteoarthritis and some of the contributory factors. He thought that several of these were usually responsible for the failure of procedures involving prostheses. He referred to the early degeneration of the joint cartilage which he ascribed mainly to alteration of joint pressures. This caused an increase of osteogenesis in the area of greatest pressure, causing bone sclerosis, and a vascular invasion of the joint cartilage in the area of less pressure, producing the osteophytes. Only procedures that allowed a nearly normal distribution of tissue pressures and so prevented these changes would have a chance of success. Finally he offered data in support of the vascular nature of the pain. He ascribed the satisfactory relief obtained at Oxford with Girdlestone's pseudarthrosis in patients in whom other methods had failed to the elimination of joint pressure.

Dr H. Debrunner (Zurich), speaking for himself as well as his Swiss colleagues, had concluded: 1) that the success of surgical treatment of arthrosis of the hip was very modest, and therefore the indications for operation demanded caution; 2) that the results of minor intervention—such as denervation, forage, etc.—were temporary and rarely better than those given by conservative measures; 3) that intertrochanteric osteotomy, more and more adopted, was a palliative operation likely above all to benefit pre-arthrosis in young people; 4) that arthrodesis was a very useful operation, but demanding precise indications and perfect technique; 5) that the orthopaedist should be familiar with all methods and adopt what suited the particular case.

Dr G. Wiberg (Lund) quoted statistics from six of the principal orthopaedic centres in Sweden, covering twenty-five years, showing that the chief operations were obturator neurectomy, subtrochanteric osteotomy (McMurray), arthrodesis and arthroplasty. Minor operations had given inconstant and temporary results. Arthrodesis, after losing ground to the acrylic prosthesis, had now regained it, and was regarded by him as the most reliable.

Professor U. Camera (Turin) reported his experience with curettage of the cancellous bone of the femoral head through a small cervical window and filling the cavity with cancellous bone, sometimes supplemented by removal of osteophytes and transposition of the greater trochanter. With this treatment, in contrast to many others, improvement usually increased with time. *Mr John Charnley* (Manchester) gave his experience of central dislocation. Even though fusion sometimes failed, the patients complained of little or no pain. The addition of temporary metallic internal fixation much increased the chances of bone fusion and dispensed with the need for plaster immobilisation and consequent knee troubles. Properly performed, the operation was safe in advanced age. *Dr J. Debeyre* (Paris) had carried out 125 Charnley " arthrodeses " with bony union in fifty-two and only fibrous union in seventy-three. Nevertheless the results were assessed as good in seventy-seven, fair in thirty-nine and poor in nine. *Dr W. Dega* (Poznan) had combined Charnley's procedure with osteotomy. *Dr A. Chakir Chakar* (Istanbul) had encountered disappointments after arthroplasties with acrylic heads, and preferred sliding osteotomies of the type of Brüeker and Moser, based on the mechanical source of the troubles, and giving very satisfactory results. He had also had encouraging results from Camera's operation. The possibilities of conservative treatment, especially pericapsular injection of analgesics must not be neglected.

Professor Bryan McFarland (Liverpool) spoke of McMurray's osteotomy. After a trial of the now popular internal fixation he had reverted to plaster. This had not unduly prolonged the convalescence, as witness a patient who had walked on board ship with a soundly united osteotomy three months after an osteotomy supplemented by a bone graft. Patients operated upon twenty-one years ago, and more, now had an average range of knee movement of 104 degrees. Immobilisation need not be very rigid if there were reasons to the contrary and one patient had been freed three weeks after operation without ill effect. Relief of pain, which was absolutely certain, was not always the consequence of displacement.

Professor R. Merle d'Aubigné (Paris) had reverted from arthroplasty to osteotomy, which gave good results, especially that of Pauwel. Charnley's operation habitually failed to give bony union and was therefore not an arthrodesis. *Dr G. Rieunau* (Toulouse) had formerly done mainly acrylic replacement of the femoral head, then mainly osteotomies and now mainly a shelf or buttressing operation (butte). *Dr P. R. Lipscombe* (Rochester, U.S.A.) referred to acetabuloplasty, forage, denervation, the operations of Collona, McMurray, Smith-Petersen, Judet, Austin-Moore, Brittain and Thompson, and to arthrodesis. He thought that osteotomy should be added to arthrodesis as the routine operation, and he also liked a supplementary iliac graft. Conservatism was not to be despised. *Dr A. S. Palazzi* (Barcelona) described an operation for transplanting a muscle flap to a cavity above the acetabulum and another to the femoral neck. This had given complete relief in 80 per cent of the patients and he had been able to return patients to work four to six weeks after operation. *Dr A. Gruca* (Varsovie) described his well-known rather elaborate osteotomy giving the results in twenty-four hips in twenty-three patients as very good six, good nine, satisfactory six, and unsatisfactory three.

Causes and prevention of congenital malformations of spine and limbs—*Drs J. Jiano, E. Repciuc, O. Vladutiu, L. Copelman, C. Bitza, M. Atanasiu* and *A. Bacauano* (Bucharest), whose paper was communicated by Professor E. Sorrel (Paris), considered irradiation of first importance, whether the influence were directly upon the gonads or through the endocrine system. They referred to toxic agents, especially alcohol, and also the indiscriminate prescription of vitamin and endocrine preparations. Other factors were infections (especially rubella), thrombosis and possibly uterine defects. *Dr M. Marcenac* (Alfort), whose paper also was communicated by Professor Sorrel, dealt with skeletal dysmorphogenesis in animals, stressing the influence on intelligence. He considered that deficiency of phosphorus, calcium, vitamin A and vitamin D deserved careful study. *Dr W. Dega* (Poznan) emphasised mechanical agents including deformities associated with placental anomalies. In particular he thought that extension was a dangerous foetal position in labour.

Mr H. J. Seddon (London) recalled that—with the increasing control of tuberculosis and the prospect of preventing poliomyelitis—congenital conditions were claiming greater attention from orthopaedic surgeons. In considering possible lines of attack, it should be borne in mind that the skeleton was not alone affected, that the information from genetics had been disappointing, and that prevention could be contemplated only with greater knowledge of causation. Of genetic or endogenous causes, he recalled such conditions as multiple exostoses, congenital dislocation of the hip and central nervous involvement. Of exogenous factors he referred to such things as irradiation, rubella and infarction. Cases of mongolism had been grouped in time and space. Endocrine effects had been studied mostly on chick embryos. The perversity of teratogenic agents was most perplexing. Possibly irradiation and vitamin deficiency were together more important than either alone. There was much circumstantial evidence that abnormal uterine compression accounted for the hyperextended knee, arthrogryphosis multiplex, congenital talipes equino varus and abnormal dimples at points of pressure; perhaps congenital uterine abnormality in the mother played a part.

Dr A. Shands (Wilmington) stated that among four million children born annually in U.S.A. the average incidence of congenital malformations was 2½ per cent, the skeletal system being affected in 25–40 per cent of cases and the neuromuscular system next most commonly. Twenty-five thousand must come yearly to orthopaedic surgeons in U.S.A. The National Institutes of Health, run by the U.S. Public Health Service, had a separate division of human embryology, and Congress had voted two million dollars for ten years for the study of cerebral malformations, especially cerebral palsy. Prospective mothers had been asked to keep a journal of possibly relevant information, which would subsequently be correlated with any congenital malformations of the child. Statistics were given of congenital deformities observed in seven years in the fifty-bed orthopaedic hospital of the Alfred I. duPont Institute.

Dr H. Idelberger (Giessen) said that a mechanical origin for congenital malformations could very rarely be conceded: those of orthopaedic interest, such as club foot, spina bifida and congenital dysplasia of the hip were genetically determined. Some countries allowed voluntary sterilisation for eugenic reasons, but there was a place besides for preventing unmanifest mutations. The danger was emphasised of radiography in pregnancy, especially of the pelvis and lumbar spine. Protection from rubella during the first three months of pregnancy was important. Reference was made also to avitaminosis, erythroblastosis and the consequences of marriages between those of Rh + or Rh − blood.

Dr de la Vega Jimena (Madrid) spoke of her experience as geneticist on the importance of intercurrent virus infection during pregnancy. The effect of external factors—toxic, physical, infectious or biological—was greatest between the second and sixth months of pregnancy.

The influence of muscle action on the apophysial and epiphysial cartilage—*Dr M. Dubois* and *Dr M. Geiser* (Berne) reported a study in rabbits of the changes in the apophysial disc of the calcaneum in response to loss of function either by plaster immobilisation or by division of the tendo calcaneus of one hind leg. In whichever way function was impaired, the changes were almost identical, the epiphysis on the experimental side becoming rapidly narrower and fusing sooner than its fellow. It seemed that what kept cartilage healthy was not just pressure but particularly intermittent pressure.

The role of compression in osteogenesis—*Drs J. Judet, R. Judet* and *J. Lagrange* (Paris) recalled that Phemister had shown the substance of a pseudarthrosis to be ossifiable and also that ossification could be promoted by a graft or by compression. Examples of tibial consolidation in two to five months were shown; but a longer, controlled, series was needed; modifications, including elastic compression, awaited investigation (and the best amount). One advantage over grafting was that only one operation was required.

Skin arthroplasty in the light of animal experiment and clinical experience—*Dr K. E. Kallio* (Helsinki) had performed arthroplasty of the hip in ninety-one cases using skin as an interposition material, with only two weeks' immobilisation; there was no infection or other complication. The skin was ultimately transformed into hyaline cartilage. At ten days granulation tissue formed between the graft and bone; at twenty days a fibrillar structure was clearly visible; at fifty days the granulation tissue was more compact; and at ninety days cartilage metaplasia had begun. By 120 days the cartilage had become hyaline, but only if there were function. It was not known yet whether this metaplasia happened in man.

The effects of intramedullary metallic nails on growing bone—*Dr F. Lee Liebolt* (New York) described experiments in puppies to determine whether intramedullary nails were as harmless in the young as in the adult. Growth was unaffected unless the epiphysial disc were transgressed, and there was no difference between the round-section Rush and the U-section Küntscher nails.

Creation of artificial tendon sheaths for tendon transfer—*Dr J. E. Milgram* (New York) had prepared a roomy bed for tendon transfer by inserting subcutaneously through a stab a polished stainless steel strip, which was removed three weeks later at the time of transfer leaving a smooth-walled tunnel for reception of the tendon. In ninety-two operations the strip had to be removed prematurely in three instances because the limb was immobilised in an unsuitable posture. The operation had proved valuable whether or not scarring was present.

Treatment of osteochondritis dissecans—*Mr I. S. Smillie* (Dundee) confirmed that separation of the fragment can be prevented by intervention and that healing can be obtained even after separation. He described fifty cases in: knee, forty-one; ankle, seven; elbow, one; hip, one. He regarded complete separation as established non-union, and fixed the fragment in its bed with a nail. Radiographic control might be necessary for finding the site to drill and was necessary after drilling. The small nail was removed at, say, eight to twelve weeks. At the ankle, the fragment had been medial in five cases and lateral in two, access being obtained by osteotomy of the malleolus. Access to the elbow was as for excision of the radius. In the femoral head retrograde bone grafting was carried out with Fitzgerald's apparatus.

Clinical and experimental use of radioactive isotopes in orthopaedics—*Dr S. von Rosen* and *Dr G. Bauer* (Malmö) said that radio-active isotopes could be used to measure rates of bone formation and bone dissolution. The daily accretion rate of calcium and the exchangeable calcium were much increased in thyrotoxicosis and depressed in myxoedema. After injection of strontium 85 (radio strontium) in a patient with a double fracture of the tibia the radioactivity of the proximal fragment was about twelve times that in the normal leg, but in the distal fragment it was about the same. A similar uptake study was being carried out with bone tumours.

Anomalies of origin, course, length and diameter of the roots of the lumbo-sacral plexus and their importance in the pathology of disc hernia—*Professor A. Bonola* (Modena) had studied, with Dr P. Bedeschi, anatomical anomalies of the lumbar roots disclosed by hemilaminectomy in more than 800 patients subjected to operation for sciatic root pain. The anomalies, which might aggravate or even determine root compression, were: 1) anomalies of origin, for instance of two roots from a common trunk; 2) anomalies of course, either "verticalisation" or "horizontalisation," with sometimes "foramen exchange," such as emergence of an unusually vertical fifth lumbar root from the first sacral foramen or an unusually horizontal first sacral root from the fifth lumbar foramen; 3) anomalies of length (brachyrhyzia); and 4) anomalies of diameter (leptorhyzia). Compression of a fifth lumbar root passing through the first sacral foramen would give the clinical features of first sacral root compression, and vice versa. In most cases in which an anatomical anomaly complicated a disc lesion, the evolution was unusually rapid, without remissions. Radiographs did not reveal any concomitant skeletal anomalies.

Two new clinical signs of lumbo-sciatica from root compression—*Dr A. Borellini* (Reggio, Emilia) described two signs present in 80 to 90 per cent of cases of lumbar or lumbo-sacral root pressure, namely: reproduction of the pain by digital pressure on the neural or vascular bundle at the fold of the groin till pulsation was obliterated or diminished; and elicitation of painful paraesthesiae by passing a current between electrodes in the fold of the groin and the lumbo-sacral region. These signs disappeared on removal of an offending disc.

Direct approaches to foci in Pott's disease—*Dr J. Debeyre* (Paris) had operated in seventy out of a hundred patients with Pott's disease undergoing antibiotic treatment. By direct access pus, caseous material and sequestra were removed. The wound was closed, a small polythene tube being left, through which streptomycin was instilled for about six weeks; antibiotics were continued up to twelve months, a plaster shell for about three to six months. Of the seventy patients, two died, three had persistent sinuses, seven were untraced and seven were too recent to assess; the others had progressed satisfactorily.

Surgical fixation of recurrent dislocation of the first cervical vertebra in children—*Dr J. T. Nicholson* (Philadelphia) described a special pattern of wiring in atlanto-axial dislocation in children with juvenile rheumatoid arthritis. For instability complicating congenital absence of the back of the atlas, reduction of the dislocation and three to six months' immobilisation in a Minerva jacket were followed by fusion of the axis to the occiput with thin tibial grafts; special caution was needed in children under four, because the occiput had not differentiated into inner and outer tables.

Treatment of slipped epiphysis and scoliosis produced in experimental animals with aminonitriles—*Dr I. V. Ponseti* (Iowa) described experiments in young rabbits and rats in which aminonitriles produced degenerative changes in the epiphysial plates leading to such changes as scoliosis and kyphoscoliosis. The effect could be suppressed by thyroxine and some of its analogues.

Possibilities of surgical intervention in the sequelae of cerebral haemorrhage—*Dr K. Jansen* (Copenhagen) discussed the possibilities of orthopaedic help based on 145 patients, 40 per cent over sixty.

Objective method for evaluation of sensibility in hand—*Dr M. E. Moberg* (Gotenberg) had increasingly supplemented the ordinary subjective tests of sensation by such functional ones as watch winding, parcel tying and doing up buttons, and by recording fingerprints on moisture-recording paper. He had used these tests for nearly two years and found them very valuable.

Results of arthroplasty of the elbow with preserved skin—*Dr V. Sanchis-Olmos* (Madrid) described skin arthroplasties carried out for tuberculous disease, hypertrophic osteoarthritis, traumatic arthritis, unreduced dislocation, osteomyelitis, etc. He particularly stressed the importance of stability. All but one of his patients were followed up for more than a year.

Bone lengthening of the femur—a new method—*Dr I. I. Alvik* (Oslo) indicated that most limb lengthening, especially in poliomyelitis, had been carried out below the knee, though often there was much shortening above. Lengthening at a bayonet osteotomy was effected by a buried screw mechanism operated by a rod projecting through the skin proximally. Four inches or more could be achieved, and the rod was then removed. Thereafter a plaster spica was applied for four to six months, followed by a short plaster spica. Some additional bone growth took place besides the lengthening obtained at operation.

Repair and reconstruction of the ligaments of the knee—*Dr J. Cabot Boix* (Barcelona) considered that the knee required more stability for sport than for work, because sport required stability in flexion as well as in extension. He considered plaster immobilisation ineffectual, however prolonged, and that early operation with complete inspection and repair of each damaged structure was essential. In sixty-four cases he had found ninety-one lesions: fifty-two in the medial ligament, twenty-nine in the anterior cruciate ligament, six in the lateral ligament, and four in the posterior cruciate ligament. He stressed the importance of general anaesthesia in early diagnosis.

The ambulant treatment of Perthes' disease—*Mr F. C. Dwyer* (Liverpool) had measured the epiphysial index at three separate stages of unilateral Perthes' disease during twelve years, and had concluded that the use of a patten-ended caliper gave as good results as methods involving prolonged recumbency.

Anomalous tendinous insertions in the pathogenesis of club foot—*Dr A. Inclán* (Havana) had confirmed anomalous tendinous insertions in a seven and a half months foetus, the subject of developmental talipes equinovarus; he had confirmed the absence of these changes in tendo calcaneus, tibialis anterior and the peronei in normal foetuses at four, seven and nine months; and he had operated in thirty-one cases (nine unilateral and twenty-two bilateral) in children of five days to eleven years of age. He concluded that anomalous insertions of the tendo calcaneus were present in all the patients and in the seven months foetus; in the other foetuses it was almost always normal. He thought that the abnormally inserted muscles provided an active force in producing deformity and should be corrected by open operation.

Longitudinal growth stimulation caused by operations for leg lengthening and osteogenesis imperfecta—*Dr H. A. Sofield* (Oak Park) had treated the grossly deformed long bones in very severe osteogenesis imperfecta by multiple osteoclasis and the insertion of intramedullary rods. The bones remained straight in spite of fractures elsewhere. In some cases a long bone was actually fragmented by open operation, and the fragments were removed and then threaded over a nail.

Decisions of Congress

In view of the serious problem presented by congenital deformities and the obscurity of their causation, the Congress recommended that its members should take the following action in their respective countries: 1) To endeavour to establish research units in appropriate centres in collaboration with other interested workers, such as obstetricians, paediatricians and embryologists; 2) To point out to their governments the social and medical importance of the problem, and to seek the financial aid necessary for the prosecution of research; and 3) When and where it seems desirable, to ask for the institution of statutory notification of disorders, such as rubella and diabetes for example, believed to be important in the etiology of congenital disorders.

At the General Assembly the president of the Society, Dr J. Delchef (Brussels) was re-elected for a further three years, and New York was unanimously chosen as the site for the next Congress in 1960. At the proposal of the U.S.A. Delegation and with the unanimous support of the Assembly, Professor J. Trueta was nominated President of the next Congress. Among the important decisions taken it was approved to elect the members of the Organising and Scientific Committees from members of the American continent, apart from the President. Professor Philip D. Wilson was elected Chairman of the Organising Committee.

ANNOUNCEMENTS

BRITISH ORTHOPAEDIC ASSOCIATION

Those wishing to present papers at the Autumn Meeting of the British Orthopaedic Association, to be held at Oxford in October 1958, should submit a précis of 400 words to the Honorary Secretary, British Orthopaedic Association, 47 Lincoln's Inn Fields, London, W.C.2, before August 1, 1958.

JOURNÉES ORTHOPÉDIQUES ET CHIRURGICALES DE BRUXELLES

Journées Orthopédiques et Chirurgicales de Bruxelles, organised by the Belgian Orthopaedic Society, the Belgian Surgical Society and the Belgian Surgical Association, will be held from May 8–14, 1958. The provisional programme of the Belgian Orthopaedic Society is as follows: **May 8. Morning**—Discussion on the treatment of recent hand injuries, opened by *Monsieur J. Lorthioir, Monsieur Evrard* and *Monsieur Vander Elst.* **Afternoon**—Short papers. **May 9. Morning**—*Mr R. G. Pulvertaft:* Tendon grafts in the hand. *Dr Jörg Bohler:* The treatment of nerve injuries in the hand and fingers, and tendon transfers for paralysis of the intrinsic muscles of the hand. *Dr Morel-Fatio:* Plastic repair after burns or wounds of the hand. **Afternoon**—Short papers. **May 10.** Symposium on the immediate treatment of road injuries. *Dr M. Belenger:* Introduction. *Professor Jean Gosset:* Resuscitation. *Professor L. Christophe:* Head injuries. *Dr Dumont:* Chest injuries. *Professor Lucien Deloyers:* Abdominal injuries. *Professor R. Merle d'Aubigné:* Injuries of the limbs and spine.

Those wishing to present short papers related to the above topics, or who require any further information, should write to the Secretary, Journées Orthopédiques et Chirurgicales de Bruxelles, Hôtel de Sociétés Scientifiques, 43 Rue des Champs-Elysées, Bruxelles 5.

The closing date for receipt of papers is March 1.

ERRATA—1) Our attention has been called to an omission in the account sent to us of the May 1957 meeting of the South-West Orthopaedic Club held at Bath (Journal of Bone and Joint Surgery, **39–B,** 789). In the description of the treatment of cavo-varus foot and club foot by Mr Dillwyn Evans, the words tendo calcaneus should be followed by the words " and perform calcaneo-cuboid arthrodesis."

2) November 1957, **39-B,** 793, last line: *for* plaster, *read* water.

Book Reviews

BONE TUMORS. General Aspects and an Analysis of 2,276 Cases. By David C. DAHLIN, M.D., Consultant, Section of Surgical Pathology, Mayo Clinic; Associate Professor of Pathology, Mayo Foundation, Rochester, Minnesota. $11\frac{1}{4} \times 9$ in. Pp. viii+224, with numerous figures. Index. 1957. Springfield, Illinois: Charles C. Thomas, Publisher. Oxford: Blackwell Scientific Publications. Price 87s. 6d.

Important as the surgeon may be as a technician in the treatment of the primary tumours of bone, his contribution to the diagnosis has become of small importance compared with that of the histologist. Even the radiologist's function may often be only that of determining the site of a tumour; for his materials are mere shadows, and they are deceptive. Nevertheless the surgeon remains the dominant individual in the team, for he must have charge of the patient and not only of *the tumour*. " To cure sometimes, to relieve often and to comfort always " is Hippocratic wisdom with particular point in malignant tumours of bone. These thoughts occurred to me when I had finished reading this admirable and detailed survey from the Mayo Clinic which is based upon the studies and records made for so many years by Henry Meyerding and continued by his followers. Dr Dahlin is a pathologist who first gathered this material together for a medical exhibition. It now forms a systematic outline of 2,276 consecutive cases, and apart from being most instructive it is a testimony to the high standards of investigation and recording carried out for so long at the Clinic.

Almost on the first page the author makes a plea for expedition in following up biopsy with definitive surgery and, very naturally, emphasises the Mayo Clinic practice of using fresh frozen tissue sections. The classification follows that of Lichtenstein, but differs in finding little etiological relation of malignant and benign tumours. In each group every aspect (clinical, radiological and pathological) is dealt with in orderly sequence and is accompanied by radiological, gross pathological and histological photographs, and by histograms and diagrams dealing with age incidence and skeletal distribution. There are very few clinical photographs. The principles of surgical and other treatments are discussed as well as prognosis. The material is presented thoughtfully and with commendable conciseness and stimulates interest by being on occasion provocative. " Enchondromas of the hands usually produce no visible swelling." " The only well documented benign chondroblastoma that has undergone malignant transformation was one that had had radiotherapy."

Of giant-cell tumour also there is a suggestion that irradiation bore a significant relation to later malignant change: nevertheless one case is reported of sarcoma developing after curettage without radiotherapy. " Curettage was followed by recurrence in more than 50 per cent "—109 cases of giant-celled tumour. " Grading of these tumours as to degree of malignancy has been of no prognostic value in the author's hands."

There were 469 osteogenic sarcomas: 19 per cent of the patients treated more than five years before survived five years, and 15 per cent of those eligible survived ten years. " Because osteogenic sarcoma is a radioresistant neoplasm, ablative surgical treatment is the procedure of choice." In low femoral sarcomas the need to study the medullary contents at the site of amputation is stressed. Perhaps implicit in this is the answer to the statement that " for reasons unknown, tumors of the tibia were twice as curable as those of the femur, the five-year survival rates being 34·6 and 17·0 per cent respectively." The author throws doubt on the grading of malignancy by the method of Broders. One is tempted to ask if the high percentage of good results is in any way related to the dependence at the Mayo Clinic upon the examination of fresh frozen tissue sections. The high malignancy of Paget sarcoma is once more noted.

Myelomas are given an important place comprising as they do in this series the largest single group of malignant neoplasms of bone. The volume is completed by a chapter on " Conditions that commonly simulate primary tumors of bone "—mostly, of course, the secondary carcinomata. Tumours of the pelvic wall present a specially interesting radiological group, and it would have been well if illustrations of carcinoma of the prostate, Paget's disease and myeloma had been included as well as the carcinoma of the breast.

It seems likely that we shall in future years see an increasing number of malignant tumours of bone. This book is one that should be readily available for reference and instruction in all departments where such cases are studied. The author and publishers are to be congratulated on the excellence of its production.—Norman CAPENER.

RHEUMATOID ARTHRITIS. By Charles L. SHORT, Walter BAUER and William E. REYNOLDS. $9\frac{1}{2} \times 5\frac{3}{4}$ in. Pp. x+480, with numerous figures and tables. Index. 1957. Harvard: University Press. London: Oxford University Press. Price 55s.

The nature of rheumatoid arthritis is very complex and its etiology is still unknown. Numerous studies based upon large series of cases have been carried out before, a notable one in Great Britain being that of Fletcher and Lewis-Faning in 1945. The latter writer also carried out a study matching patients with controls in a considerable investigation sponsored by the Empire Rheumatism Council in 1950. The present study is an altogether admirable development of this statistical approach. It is divided into forty-five chapters. The first, giving a review of previous work, is followed by a discussion of the diagnostic criteria for the selection of patients. The series used was drawn from 300 consecutive patients admitted to the medical wards of the Massachusetts General Hospital between 1930 and 1936 and in whom the diagnosis of rheumatoid arthritis was supported by extensive clinical and laboratory study. The differentiation of rheumatoid arthritis from other forms of chronic joint disease is beautifully presented. The remaining five-sixths of the volume deals with the various sections of investigation such as sex distribution; age of onset; family history; allergic diseases; occupation; the menopause; precipitating factors; joints first involved; constitutional, vasomotor, neurologic and gastro-intestinal factors; lymph nodes; the skin; nodules; joint examinations; foci of infection and the pathological investigations, etc. The final chapter is a careful summary of the results. Each chapter also is provided with a concise summary and includes a brief list of objectives for further study. It is not possible to select elements of this review for special comment: every chapter contains material rich in value and, although strung upon great detail, is presented in a thoughtful and most readable style. Of some interest is the literary and typographic presentation. Each paragraph is numbered in reference to the chapter number, e.g., the eighth paragraph of the forty-second chapter, which is on *Spondylitis*, deals with " objectives for further study " and is preceded by the bold **42.8** which makes for easy reference and indexing. Incidentally this paragraph includes a question " whether or not rheumatoid spondylitis constitutes a distinct disease entity "; the answer reflects the general American tendency to regard it as not so, yet maintains an open mind. This volume is produced under the auspices of the Commonwealth Fund (of Massachusetts). Amongst the supporters of the researchers who are acknowledged by the authors, it is pleasant to see the name of one of the pioneers of modern American orthopaedics in the " Robert W. Lovett Memorial Foundation " (established by Dr Frank Ober). One cannot speak too highly of this most instructive, well written and excellently produced work.—Norman CAPENER.

ORTHOPEDIC SURGERY IN INFANCY AND CHILDHOOD. By Albert Barnett FERGUSON, Jun., B.A., M.D., Silver Associate Professor Orthopedic Surgery, University of Pittsburg; Chief of Orthopedic Service, University of Pittsburg Medical Center; with four collaborators. $10\frac{1}{4} \times 7\frac{1}{4}$ in. Pp. xii+508, with 504 figures. Index. 1957. London: Baillière, Tindall and Cox. Price £6.

The orthopaedic surgeon who would search for guidance in the management of one of those conditions from which his title derives must too often feel the temper of his enthusiasm failing as he searches journal and text for a reasoned and brief account of the source of his interest.

The author of this book recognises this paradox in his preface and with the aid of four distinguished contributors sets himself the task of making good this deficiency in the literature of our speciality. The book, which is largely written by Ferguson himself, goes a long way towards fulfilling this aim. The opinions expressed and the practice advocated rarely differ from those customary in the United Kingdom. There are differences, of course, and some, usually minor and involving degrees of emphasis rather than matters of principle, will be mentioned in this review. The illustrations are admirable, and not only clarify but amplify the text, which seldom lapses from either literacy (" institutionalisation ") or accuracy (" Dennis Browne "). The bibliography, brief and well chosen, provides an introduction to further reading when considerations of space or balance have curtailed discussion.

The first two chapters deal with disorders of the foot and knee in an orthodox manner. Many surgeons, however, might hesitate to forbid weight bearing in Köhler's disease until the bone density is repaired. The regional approach continues with a chapter on diseases of the hip joint in which the treatment of congenital disorders is discussed with admirable clarity and conviction. Less conviction is shown, perhaps justifiably, when the management of Legg-Perthes' disease is considered. The reader is urged to drain suppurative arthritis of the infant's hip as soon as the diagnosis is made—a recommendation that will be supported by many British surgeons. The decline of Pott's disease is rightly reflected by the comparative brevity of the section devoted to it. Late spinal fusion is advocated in all cases, but the possibility of direct surgical attack on the vertebral bodies with drainage is not

mentioned. Paraplegia receives scant consideration; conservative treatment, drainage and (surprisingly) spinal fusion alone are advised but antero-lateral decompression is ignored. Donaldson, writing on scoliosis, gives a good general account of etiology and management but omits any reference to prognosis in idiopathic scoliosis in relation to curve patterns. He reviews affections of the neck and introduces "congenital nuchal rigidity," a syndrome recognised by the writer of this review only after reading this book. The same author is surely a little prejudiced against Still's disease when he suggests that it is *usually* a "generalised, debilitating, malignant, ankylosing and desperate affliction—one of the unhappiest of childhood afflictions."

Stelling subscribes a chapter on the surgery of the upper limbs notable for its emphasis on congenital abnormalities and for its cautious approach to the surgical treatment of the hand crippled by cerebral palsy. In poliomyelitis he warns against the dangers of ill-considered wrist fusion and gives a concise account of the indications for and the technique of tendon transplantation and tenodesis. Albert Ferguson, Senior, describes the general affections of the skeleton, and, although he uses a terminology which may sometimes be unfamiliar to British readers, his classification based on his concept of the underlying defect is helpful in so far as it allows him to forego the tedious catalogue that so often causes this chapter to remain unread in other text-books. Disorders of calcium metabolism are described by Klein, whose achievement it is to simplify the complex chemical problems that characterise those diseases without once appearing to patronise those among his readers whose tenuous grasp of biochemistry was totally relaxed when mille-equivalents replaced milligrams.

The concluding chapters on affections of muscle, the nervous system (including poliomyelitis) and tumours of bone are worthy if pedestrian. Nevertheless those practising surgery in cerebral palsy cannot ignore the warnings contained in this section and must applaud the author's discrimination in excluding those operations which though often appearing in the abstract as often appear irrational in the presence of a cerebral cripple. Leukaemia, often and perhaps increasingly with us, is not mentioned in the last chapter on malignant tumours, which is a pity because, with the bone changes found in neuroblastoma, this disease can be both deceiving and humiliating.

This book whilst filling a hiatus is more than a stop-gap. It is orthodox without being pedantic. It is expensive but worth it.—G. C. LLOYD-ROBERTS.

L'ARTHROGRAPHIE OPAQUE DU GENOU. Contribution au Diagnostic des Dérangements internes du Genou. By Paul FICAT, Assistant de Chirurgie des Hôpitaux, Diplômé d'Électro-Radiologie. Preface by Professor R. MERLE D'AUBIGNÉ. $9\frac{1}{2} \times 6\frac{1}{2}$ in. Pp. vi+243, with 139 figures. 1957. Paris: Masson & Cie, Editeurs. Price 3,800 fr.

This book can be read with advantage by anyone interested in lesions of the knee, whether or not he is interested in arthrography. As the author implies, it is only by arthrography that a true interpretation of the anatomy of the joint can be obtained when the structures are undisturbed by dissection either in the anatomy department or the operation theatre. By no means confined to diagnosis, the work is divided into three main sections, the first on technique, the second on the normal joint, and the third on the abnormal joint.

The technique, using an opaque iodine compound, is based on that described by Lindblom. It involves five routine views, all showing each meniscus tangentially. Extra views, such as the "skyline" view of the patella, are used when occasion arises.

The book is beautifully illustrated throughout. In 230 pages there are 139 illustrations, most of which occupy a whole page and consist of an arthrogram and explanatory diagram. The description of the normal joint leaves no nook or cranny unexplored and no shadow unexplained. In this, the book has its strongest and its weakest points. Where so many shadows are present, it might perhaps be better to emphasise the salient features at the expense of the others.

The section on pathological lesions describes not only the arthrographic appearance of the usual traumatic lesions of the meniscus and articular cartilage, but gives also a more speculative description of the appearance in arthritis, whether traumatic, metabolic or infective. All the usual meniscus lesions are illustrated, and the results are given. The number of lesions diagnosed accords with the experience of others, but it is a little disappointing that no attempt is made to indicate the value of a negative diagnosis, which can be as great as a positive. The more speculative discourse on arthritis is of interest rather than of true diagnostic value, but provides much food for thought about the local progressive pathology.

It is unlikely that anyone reading this book critically—and it must be read critically—will agree with all that is said; but it must be admitted that even the most controversial points are not merely stated but clearly illustrated.—E. W. SOMERVILLE.

RECONSTRUCTION SURGERY AND TRAUMATOLOGY. Edited by M. LANGE and twenty-one other editors. Volume IV. $9\frac{3}{4} \times 7$ in. Pp. iv + 222, with 201 figures. 1957. Basel/New York: S. Karger. Price S.Fr. 46.

This volume, like volumes I, II and III (published in 1953, 1954 and 1956), is concerned exclusively with problems of trauma. There are two articles written in English, one in French and the rest in German. The first part of Volume IV is devoted to three articles by well-known authorities on recurrent dislocation of the shoulder. Eyre-Brook (Bristol) states clearly the indications for the operation of Bankart and the Putti-Platt procedure. Both of these operations are described in detail by Scaglietti and Calandriello (Florence). Lange (Munich) prefers an operation which includes an anterior bone graft and shortening of the subscapularis. Fineschi (Florence) and Böhler (Linz) contribute interesting articles on the late results of traumatic dislocation of the hip. Debrunner (Zurich) recounts his experiences with recurrent dislocation of the patella. Jonasch (Vienna) reports on more than one thousand tears of ligaments of the knee and concludes that conservative treatment is better than treatment by operation, whereas Merle d'Aubigné (Paris) after consideration of fifty-five cases is in favour of operative repair. Palmer (Stockholm) gives clear evidence of the good results to be obtained by early diagnosis and early repair by operation. Lange (Munich) also advocates operative intervention and gives the indications for repair in the early and late stages. This is the best volume of the series and no surgeon who deals with knee injuries can afford to ignore the excellent reports by these continental authors.—L. W. PLEWES.

ARTIFICIAL LIMBS. By Leon GILLIS, *M.B.E.*, M.B., M.Ch.Orth., F.R.C.S., Consultant Orthopaedic Surgeon, Queen Mary's (Roehampton) Hospital, London. With a foreword by Sir Harry PLATT, LL.D., M.D., M.S., F.R.C.S., F.A.C.S.(Hon.), President, Royal College of Surgeons. $10 \times 7\frac{3}{4}$ in. Pp. xiv + 449, with 295 figures and 6 plates. 1957. London: Pitman Medical Publishing Co. Ltd. Price £15.

Mr Gillis occupies a unique position as Orthopaedic Surgeon to Queen Mary's Hospital, Roehampton, for there he has not only the surgical care of a large and continuous flow of amputation cases but also at his elbow, the country's main limb-fitting centre, only limb research centre and largest limb factory. He is therefore ideally placed to produce an authoritative text-book on artificial limbs. By writing *Artificial Limbs* he has shown that he also has the enthusiasm and energy. Unfortunately, however, experience, enthusiasm and energy are not necessarily synonymous with successful authorship, and it seems very doubtful whether this book will achieve the status that all would have hoped for it. The work is divided into three main parts: Limb-fitting, Artificial Limbs, and Rehabilitation. The information contained therein is very extensive and valuable, and in case there are any gaps, which seems doubtful, there are exhaustive lists of world literature at the end of every chapter. But, if a book aspires to become a standard text-book, it requires a great deal more accuracy of detail, clarity of thought and exposition, rationality of sequence and relevance of comment than is apparent here. Though clearly an enormous amount of work has gone into the preparation of this volume, it is difficult to see how the prohibitive price can be justified, particularly as the blocks of over a fifth of the illustrations were available from *Amputations*, the author's previous publication. With the best will in the world, one cannot predict a ready sale for *Artificial Limbs*.—F. G. St Clair STRANGE.